HEALTH CARE SERVICE UTILIZATION AND MEDICAID COSTS AMONG CHILDEN WITH AND WITHOUT OROFACIAL CLEFTS IN NORTH CAROLINA, 1995-2002

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ABSTRACT

CYNTHIA HOUSTON CASSELL: Health Care Service Utilization and Medicaid Costs among Children With and Without Orofacial Clefts in North Carolina, 1995-2002
(Under the direction of Julie Daniels)

Orofacial clefts (OFC) affect about 6,800 children annually in the U.S. and one in 890 children in North Carolina. Previous studies on health service utilization and cost among children with special health care needs are limited. In 1993, the American Cleft Palate-Craniofacial Association (ACPA) developed treatment recommendations for patients with OFC. No study has examined the timeliness of services according to these guidelines.

North Carolina vital statistics, birth defects registry, and Medicaid enrollment and paid claims were matched to identify resident children born 1995-2002 with (cases) and without (controls) OFC who were continuously enrolled in Medicaid. Average number of paid claims and cost per child with and without OFC were determined for medical, inpatient, outpatient, dental, well-child care, mental and home health. To determine the effect of maternal, child, and system characteristics on health service use and cost, Poisson regression and two-part modeling were employed. Multivariate logistic regression was used to examine factors associated with timely receipt of cleft surgery.

Children with OFC utilized significantly more health services and had significantly greater costs than unaffected children during infancy. Total cost for children with OFC was $12,792,634 compared to $2,212,839 for unaffected children on Medicaid. Generally, characteristics associated with health service use and cost varied among categories and cases and controls. Common characteristics associated with greater service use and cost included

being born low birth weight, residing in the northeastern and western regions, and receiving maternity care coordination (MCC).

About 78% of children with OFC had primary cleft surgery, 51% received speech and language therapy, and fewer than 28% received other specialized services within ACPA recommendations. Timely receipt of cleft surgery varied significantly by receipt of MCC, prenatal care source, and perinatal care region.

This study confirms children with OFC have significantly greater health service use and costs than unaffected children. Most children with OFC received cleft surgery, yet many did not receive specialized services within ACPA guidelines. To improve health outcomes, efforts need to be addressed to increase timely receipt of services. These results can inform policy, program development, and service planning and delivery in the state and in the U.S.

This dissertation is dedicated to relatives who died during my time as a graduate student in the Maternal and Child Health Department, including Bill Banister, my uncle, Mildred Houston, my grandmother, Douglas Wolf, my husband's grandfather, and Mary Wolf, my husband's grandmother. It is also dedicated to the most loyal basset hound, Goldie, who I got as a puppy when I started graduate school and died a little over a year ago. I am confident she is still with me through my now one year old basset hound, Chloe.

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LIST OF ABBREVIATIONS

ACPA	American Cleft Palate-Craniofacial Association
CI	Confidence interval
CL	Cleft lip
CLP	Cleft lip with cleft palate
CP	Cleft palate
CSHCN	Children with special health care needs
EPSDT	Early and Periodic Screening, Diagnostic, and Treatment
GIS	Geographic information system
LBW	Low birth weight
MCC	Maternity care coordination
NCBDMP	North Carolina Birth Defects Monitoring Program
OFC	Orofacial clefts
OR	Odds ratio
PNC	Prenatal care
WIC	Special Supplemental Nutrition Program for Women, Infants, and Children

CHAPTER I

INTRODUCTION

Statement of the Problem

Orofacial clefts (OFC), which include cleft lip (CL) with and without cleft palate and cleft palate (CP) alone, are the third most common birth defect in the United States (1, 2). On average, 20 infants a day are born with OFC and affect about one in every 700-1,000 infants (3). Currently, OFC affect about 6,800 children each year in the United States (2, 3). In North Carolina, approximately 180 children per year are diagnosed with an orofacial cleft (4). The prevalence of OFC was 15.3 per 10,000 live births for 1995-2002 in North Carolina. The prevalence of children with CL with and without CP was 9.6 per 10,000 live births and 6.3 per 10,000 live births for children with CP for 1995-2000 in North Carolina (5).

Orofacial clefts can impair the development of teeth, speech, hearing, and feeding capabilities and often impair psychomotor and cognitive skills, thereby creating physical and emotional stress for children and their families (4, 6-17). Such problems can result in significant direct and indirect costs. Direct costs include costs to the health care system itself and indirect costs include costs related to mortality, morbidity, and disability. In the United States, in 1992 dollars, the average lifetime cost per child with CL or CP was estimated at $101,000 for a 1988 birth cohort (18). For the same time period in the United States, the total costs of children with OFC was estimated to be $697,000,000 (16). In North Carolina for a 1988 birth cohort in 1992 dollars, the total lifetime cost per child with CL or CP was estimated at $16,171,027 (18).

Due to the prevalence and high cost of children with birth defects such as OFC, improving the health of these children is an important public health goal. However, there is a paucity of current information on health care service utilization and costs for children with OFC and other birth defects. National data that pertain to children with special health care needs (CSHCN) and selected subgroups of this population, such as children with asthma, fetal alcohol syndrome, cerebral palsy, and developmental disabilities, are limited and do not specifically address children with OFC (16, 18-26). For instance, the recent National Survey of Children with Special Health Care Needs provides some state-specific information related to prevalence, services, and health care access for CSHCN, but key demographic and county level data are lacking (27-30). Moreover, few data are available to describe variability in service use and cost by child characteristics such as age and diagnosis. These factors are critical because patterns of medical and health-related service use and costs for children with birth defects can vary considerably due to biological, familial, social, and developmental factors (20, 31).

There are no recent data on the effect that geographic barriers, such as distance traveled to and from specialized health care providers, have on service utilization and timely receipt of services. To date, there has only been one study that examined distances to health care providers among children with OFC. However, this study was conducted in the late 1960's, which was prior to the availability of advanced geographic information system (GIS) analysis. This study also failed to control for important demographic variables (32).

Components of health care service utilization and cost for children with birth defects such as OFC have not been well studied. In addition, an overall understanding of health service utilization and cost patterns among different age groups for children with OFC is lacking.

Such an understanding is critical for developing cost-effective strategies for prevention of OFC as well as receipt of and, moreover, timeliness of services for this population. These gaps in the literature were recently identified as research priority areas by the National Center on Birth Defects and Developmental Disabilities, Centers for Disease Control and Prevention (33). This information is important for enhancing the long-term health and quality of life for children with OFC and their families.

Specific Aims

Aim 1: To describe health care service use and costs among children with OFC (cases) and without OFC (controls) on Medicaid during the first five years of life.

1. What are the differences in specific health care service use and cost by service type such as medical, inpatient/hospitalization, outpatient, mental health, home health, dental, and well-child care between cases and controls for each year of life?
2. What are the differences in cumulative health service use and cost between cases and controls for each year of life?
3. Among cases, what are the differences in specific health care service use and cost by cleft type and presence of other anomalies for each year of life?

Aim 2: To determine the effect of selected individual (maternal and child) and system characteristics on health service use and costs among children with OFC and unaffected children on Medicaid during the first year of life.

1. What maternal, child, and system factors are associated with medical, inpatient, outpatient, mental health, home health, and total service use among cases and controls?
2. What maternal, child, and system factors are associated with medical, inpatient, outpatient, mental health, home health, and total costs among cases and controls?
3. Among cases, what is the effect of cleft type and presence of other anomalies on health service utilization and cost?

Aim 3: To assess the timeliness of certain services according to the ACPA recommendations among children with OFC on Medicaid.

1. What proportion of children with cleft lip (CL) received primary cleft surgery within the first six months of life?
2. What proportion of children with cleft palate (CP) or cleft lip with cleft palate (CLP) received primary cleft surgery within the first 18 months of life?
3. What maternal, child, and system characteristics are associated with timely receipt of primary cleft surgery?
4. What proportion of children with OFC received audiological and dental services and speech and language therapy within the first year of life?
5. What proportion of children with OFC received otolaryngologic care within the first six months of life?
6. What proportion of children with OFC received genetic services within the first two years of life?

Background

Definition and Epidemiological Profile of Birth Defects in the United States and North Carolina

As defined by the March of Dimes, a birth defect is an abnormality of function, structure, or body metabolism that is present at birth and results in mental and/or physical disability (34). A major defect is a defect of cosmetic or functional significance requiring some medical intervention whereas a minor defect is a defect that is of minimal or no functional or cosmetic significance. Minor defects occur in fewer than five percent of the population (35). Annually, 120,000 infants are born with a major and/or minor birth defect in the United States, which translates to one in 33 infants (34, 36, 37).

In the United States, birth defects remain a leading cause of infant mortality, accounting for greater than 20% of all infant deaths (36-40). In 1997, congenital malformations accounted for 22.1% of all infant deaths compared with 15.1% in 1970 (36). In 1999, birth defects accounted for about one in five infant deaths in the United States (41).

In North Carolina, more than 3,500 children, which is about one in 33, are born each year with a serious birth defect (4, 36, 42). This figure represents about three percent of all live births in the state (42). In 2003, 27.2% of children with birth defects died within the first year of life (42).

Children with birth defects have special health care needs and are usually at an elevated risk for a chronic developmental, physical, emotional, or behavioral condition. Because children with birth defects generally need health and related services beyond those required by children in general, children with birth defects represent a subset of CSHCN.

Orofacial clefts occur when the structures of the mouth fail to develop correctly during the third and twelfth weeks of pregnancy (4, 43). Because of the physical nature of OFC, these

types of birth defects can be readily diagnosed at birth and therefore are among the most frequently and readily diagnosed major birth defects (1, 44). This group of birth defects includes CL with and without CP and CP alone (35). Cleft lip with or without CP occurs in the primary palate and/or lip that form between the third and seventh weeks of gestation. Cleft lip with or without CP occurs in 1 in every 1,000 infants born. In comparison, CP occurs in the secondary palate that develops between the fifth to twelfth weeks of gestation and occurs less frequently in about 1 in every 2,000 births (35, 43, 45, 46).

Orofacial clefts that occur with other birth defects have different epidemiologic characteristics compared to those clefts that occur alone. Consequently, OFC can be further divided into "isolated" (non-syndromic) clefts, which is defined as clefts that occur alone or with minor defects and "non-isolated" (syndromic) clefts, which are clefts that are associated and/or occur with other major defects (35, 46). Some researchers use isolated and non-syndromic and non-isolated and syndromic interchangeably while other researchers define isolated clefts as clefts that occur with no other defects.

There are about 400 syndromes that have some form of OFC along with other birth defects (43). Several studies have examined the percentage of syndromic and non-syndromic CL with or without CP and CP. In general, non-syndromic CL with or without CP account for 70-80% of all OFC cases (3, 35, 40, 47, 48). The most common minor anomaly in patients with non-syndromic CL with or without CP is hypertelorism, which is an abnormal distance between two organs (35). In contrast, CP is much more likely to be syndromic and associated with other defects (35, 47-49). In previous studies, the incidence of non-syndromic CP has ranged from 41% to 55% (35, 48). The most common syndrome associated with CP is Stickler's syndrome, which has been identified as the most common

syndrome causing Robin sequence (35, 50). Stickler's syndrome is a hereditary progressive arthro-opthalmopathy, which is an association of degenerative joint disease and eye disease. Other anomalies known to be associated with CP include abnormally wide distance between the eyes, defects of the abdominal wall, conotruncal heart defects, dental malformations, and underdevelopment of the facial bones (17, 49, 51-55). Complex syndromes associated with OFC may not manifest until later in life.

Differentiating between non-syndromic/isolated and syndromic/non-isolated OFC is important because such a distinction can ultimately affect timely receipt of services, health service utilization, and thereby costs. In this dissertation, results were stratified by cleft type and presence of other anomalies to examine such differences in children with OFC. Cleft type was defined as CL alone, CP alone, and cleft lip with cleft palate (CLP). Children with isolated clefts were defined as children with OFC that occurred alone with no other associated syndrome and/or birth defect. Children with non-isolated OFC were defined as children with OFC that occurred with an associated syndrome and/or other birth defect.

The prevalence of OFC can vary according to maternal age, gender, race/ethnicity, socioeconomic factors, and maternal prenatal exposures such as smoking, alcohol, and vitamin deficiency (35, 46, 56-60). The prevalence of children with OFC is higher among younger mothers, mothers of Asian and American Indian descent, mothers who smoke during pregnancy, and/or mothers who are not married. Specifically, Native Americans have the highest prevalence of CL with or without CP with 3.6 cases per 1,000 births, followed by Asians with 2.1 cases per 1,000 Japanese live births and 1.7 cases per 1,000 Chinese live births (35, 43, 61). An increased prevalence also exists among children with non-isolated CP. Male children are affected more frequently by non-isolated CL with or without CP than

female children, and female children are affected more frequently by isolated CP than male children. The prevalence of children with OFC is also higher among families with low socioeconomic status (35, 43, 46, 47, 50, 56, 62, 63).

Despite the identification of certain environmental, demographic, and genetic risk factors associated with OFC, the etiologic factors identified by previous studies are inconsistent due to differences in study design and population studied. Potential biases in these previous studies include small sample size, lack of population-based data, study of combined instead of separate phenotypic groups, reliance on birth certificate reporting of OFC, and omission of differences between non-isolated and isolated clefts (46, 64).

Approximately 180 children per year are diagnosed with OFC in North Carolina (4, 42). For 1995-2002, the prevalence of OFC was 15.3 per 10,000 live births. For 1995-2000 in North Carolina, the prevalence of CL with and without CP was 9.6 per 10,000 live births and 6.3 per 10,000 live births for CP alone (5). In North Carolina, CL with or without CP is more common among Whites and in males. For 1995-1997, the western, northwestern, and northeastern regions had higher rates of OFC than the eastern, southwestern, and southeastern parts of the state (4).

Health Service Use and Cost among Children with Special Health Care Needs

Several studies have examined health service utilization and costs among children with and without special health care needs such as children with attention deficit hyperactivity disorder, asthma, and mental health conditions (20, 21, 26, 30, 65-95). It is well documented that CSHCN utilize services more and incur greater health care costs than children without special health care needs. In general, previous studies have demonstrated that service use and

costs for CSHCN vary considerably across different chronic condition categories and across different categories such as inpatient, physician, and outpatient services (20, 21, 70).

Previous studies on CSHCN have also shown disparities in access and service utilization for children with and without special health care needs (26, 88, 96-100). These studies have clearly demonstrated minority children have greater difficulty in accessing health care and have poorer health status (78, 90). Studies examining CSHCN indicate that minority CSHCN experience differences in insurance coverage and utilization of health services (26, 88, 95-98). In addition, these studies illustrate Black CSHCN appear to rely more on the various public insurance programs whereas Hispanic children are twice as likely to be uninsured compared to White CSHCN (83, 96). There are also differences in utilization patterns, with White CSHCN using ambulatory services more often while minority CSHCN rely on emergency rooms for their regular source of care (97). In addition, CSHCN from families with low education attainment and non-Hispanic Black CSHCN are significantly less likely to use many medical and health-related services (26).

Recent data on health care utilization are lacking for children with birth defects, especially for children with OFC, in North Carolina. National data that pertain to CSHCN and selected subgroups of this population, such as children with chronic conditions like asthma, fetal alcohol syndrome, cerebral palsy, and developmental disabilities like ADHD, are limited (16, 18-26, 77). The recent National Survey of Children with Special Health Care Needs provides state-specific prevalence estimates of CSHCN, describes the types of services these children need and use, and examines access to care for CSHCN (27-30, 93, 95). However, data on county-level, demographics such as child's age, and specific chronic conditions like birth defects are lacking from this national survey. These data are critical because patterns of

medical and health-related service use and expenditures for children with birth defects can differ considerably due to biological, familial, social, and developmental factors (20, 31). Similarly, few studies are available that describe determinants of medical and health-related service use among subgroups of CSHCN.

Several other national datasets such as the Healthcare Cost and Utilization Project and Medical Expenditure Panel Survey have been employed to analyze expenditures among CSHCN. However, most studies did not adequately control for factors that could have influenced utilization and thereby costs. These factors include maternal age, education, race/ethnicity, marital status, birth hospital, presence of birth defects, and place of residence. Some of these previous studies employed Medicaid paid claims to analyze health service use and expenditures. However, the authors did not consider the length of enrollment in Medicaid. If Medicaid enrollment is ignored, then the results are not an accurate representation of the true effect measure over time, thus masking significant associations. Most importantly, previous studies on expenditures among CSHCN did not focus on children with birth defects and did not verify the condition (20, 70, 101).

Health Service Use and Cost among Children with Birth Defects

Children with birth defects, including OFC, remain the leading cause of pediatric hospitalizations and expenditures in the United States. In general, approximately 25% - 30% of pediatric hospital admissions are due to children with birth defects (37). In the United States each year, the total costs for the hospital care of children with birth defects exceeds $1.4 billion (37). Two recent studies were conducted on service use and cost of birth defects using the Healthcare Cost and Utilization Project data (102, 103). Russo et al. found that in

the United States in 2004, children with birth defects totaled $2.6 billion in hospital costs, which was much higher than past cost estimates (103).

In North Carolina in 2000, 30% of admissions to pediatric hospitals resulted from birth defects (4). In 2000, children less than 18 years old with a birth defect accounted for more than $53.1 million total inpatient hospital charges in the state (4). These charges excluded physician costs, medication, and other services (4). In 2003, inpatient hospital care for children with birth defects less than 18 years old was $73.4 million. In North Carolina, the total cost to Medicaid for children with birth defects in the first year of life was $65.1 million in 2003 (42).

Only two recent studies have examined health service use and costs among children with birth defects, including OFC. These studies used Healthcare Cost and Utilization Project data from 2003 and 2004, and examined hospitalizations and costs from hospital discharge data (102, 103). Russo et al. found that in 2004 in the U.S., children with birth defects had more than 139,000 hospital stays due to their birth defects treatments, which represented a hospitalization rate of 47.4 stays per 100,000 total children. Hospitalizations due to birth defects were longer and occurred more frequently among males and younger children compared to hospitalizations for all other non-obstetrical conditions (103). For children with CP, the total number of hospital stays was 2,900 and the mean length of stay was 2.1 days. In comparison, children with CL with or without CP had a total of 4,900 hospital stays and the mean length of stay was 1.9 days (103). Robbins et al. found that in 2003, 2,187 [95% confidence interval (CI): 2,109-2,264] hospitalizations were due to newborns with isolated CP and 3,496 (95% CI: 3,386-3,585) hospitalizations were due to newborns with CL with or without CP. In addition, the authors found that newborns with CP alone averaged 10.2 days

(95% CI: 9.6-10.9) and newborns with CL with or without CP averaged 5.6 days (95% CI: 5.3-5.9) in the hospital (102).

Children with OFC can incur significant direct and indirect expenditures. Direct expenditures include expenditures to the health care system and indirect expenditures include those associated with mortality, morbidity, and disability. These expenditures can be due to the impairment of teeth, speech, hearing, feeding, and psychomotor and cognitive skills (4, 6-17). In 1992 dollars, Waitzman et al. calculated the average lifetime cost per child with CL or CP was $101,000 and the overall total costs of OFC was $697,000,000 for a 1988 birth cohort in California (16, 18). Harris and James used the data from California to estimate state-by-state cost of birth defects in 1992 U.S. dollars and found the total lifetime cost for all children with CL or CP was $16,171,027 in North Carolina (18).

The studies conducted by Waitzman et al. in 1994 and Harris and James in 1997 are the two most comprehensive studies conducted on costs of birth defects (16-19). A major strength of the study by Waitzman et al. was that they employed several major data sources to determine costs such as the California Birth Defects Monitoring Program, National Health Interview Survey, California Office of Statewide Health Planning and Development Hospital Discharge Abstracts, California Department of Developmental Services, National Longitudinal Study of Special Education Students, California Special Education Expenditure, and California Age-Sex Earnings Profiles. Consequently, costs associated with mortality, morbidity, including disability (indirect costs) and developmental services and medical costs, including inpatient, outpatient, and long-term care (direct costs) were included in the analysis. This study also adjusted for costs incurred in the future such as potential reduction in salaries by including a discount rate of 2-10%. Costs were estimated accordingly over a

five-year period (1988-1992) among children with the selected birth defects. Another strength of this study was that an incidence approach under cost-of-illness methodology was utilized to analyze the costs. Cost-of-illness methodology generally includes an incidence and/or prevalence approach. An incidence approach includes a per person cost at each stage of the condition and is usually utilized to estimate cost-effectiveness analyses of prevention strategies such as intake of folic acid and prevention of neural tube defects. In comparison, a prevalence approach is more appropriate for examining treatment strategies (16, 17, 19). The study conducted by Harris and James in 1997 improved the study by Waitzman et al. by including data from the Metropolitan Atlanta Congenital Defects Program. Cost estimates were made for each state for each of the selected 18 birth defects, including OFC (18).

The studies by Waitzman et al. and Harris and James had regional limitations and are now outdated. In addition, they do not provide information by cleft type or by other types of defects such as isolated or non-isolated OFC (16-19). As described earlier, different cleft types require different services and frequency of services, which ultimately affect the cost of these defects. By stratifying by cleft type, one has a better understanding of the costs affiliated with non-isolated and isolated clefts and CL compared to CP.

To determine costs of children with OFC, Berk et al. examined patients from a 1994 birth cohort seen at the University of Pittsburgh Cleft Palate-Craniofacial Center. The authors stratified by cleft type such as CL, CP, and CLP and calculated the minimum, maximum, and average number of services received over the first five years of life. The authors found that the minimum, maximum, and average fees billed were higher for CLP than for CL alone and CP alone over the first five years of life. For example, for CLP, the average fees billed for the

Cleft Palate-Craniofacial Center services and plastic surgery was about $10,329 compared to $3,610 for CP alone (31).

Despite these results, there were several severe limitations to this study such as the small sample size (N=38) and selection bias due to sampling from one clinic. Furthermore, the costs examined in this study were fees or charges billed to patients and not the actual amount paid or collected for the services. Charges are not good proxies for costs because charges cover costs of indigent and unreimbursed care, community service, and capital expenses. Additionally, they are adjusted based on negotiated contracts and community practices and are usually inflated cost estimates (77, 104).

A recent study examined the cost of medically treated craniofacial conditions, which included children with OFC. The authors found that in 1999, the estimated inpatient and outpatient annual cost of craniofacial congenital conditions in the U.S. was over one billion dollars. However, this study grouped OFC into all craniofacial congenital conditions, so there was no disaggregation by type of malformation or age-specific data. Additionally, the authors utilized administrative data only and did not verify the condition (105).

Recent studies by Russo et al. and Robbins et al. were conducted on hospitalizations and costs of children with birth defects, which included OFC. In the study that examined hospitalizations in 2004 for children with birth defects, the mean cost of isolated CP was $5,400 and the aggregate cost for this cleft type was $15,506,700. The mean cost of CL with or without CP ($5,500) was similar to isolated CP; however, the aggregate cost for CL with or without CP was $27,155,800 (103). Robbins et al. examined hospital costs during the newborn period and found the mean hospital charge for CP alone was $33,387 and total

hospital charges were $72,914,132. For CL with or without CP, the mean hospital charge was $15,397 and total hospital charges were $53,630,046, lower than for CL (102).

The major strength of these studies was providing updated information on costs of children with birth defects. However, these studies suffered from several limitations, such as excluding physician / professional fees. Additionally, the unit of analysis was the hospital discharge, not the child. The authors only examined one health service use and cost category (hospitalizations) and did not control for any factors that might be associated with health service use and cost such as child's age and presence of other anomalies. One of the studies only examined inpatient use and costs in the newborn period, which does not represent all the service use and cost of children with OFC in a given year, which is important when planning services and treatment. In addition, the authors examined charges rather than costs, which do not represent what the actual reimbursement was for services rendered for these children. Lastly, the authors only stratified by CP alone and CL with or without CP, which masks any effects of isolated CL, and they did not examine the presence of other birth defects.

Other studies have been conducted that evaluated costs of children with OFC. However, these studies examined only certain aspects of treatment like nasoalveolar molding, gingivoperiosteoplasty, and alveolar bone graft, and/or specifically addressed one type of cleft type such as unilateral cleft alveolus or complete unilateral CLP (25, 106, 107). In addition, these studies did not analyze predictors of health service utilization or costs among children with OFC.

Recent data on health service use and expenditures are lacking for children with birth defects, especially for children with OFC in North Carolina. Previous studies have been restricted to certain periods of time, such as the newborn period, and did not analyze service

use and cost per child with OFC. In addition, the most comprehensive cost estimates are outdated and do not consider health service use and costs of isolated CL and CLP separately (16, 18-26). Few data are available to describe variability in service use and cost by service category and by maternal, child, and system characteristics. These factors are critical because patterns of medical and health-related service use and expenditures for children with birth defects can differ considerably by birth defect type (20, 31). Understanding patterns of health service use and expenditures can better help program and policy makers target populations in need of services and target areas for reducing costs and appropriately allocating health care resources.

Recommended Services and Treatment for Children with Orofacial Clefts

Recommended services and treatments exist for children with an array of selected birth defects such as spina bifida, a type of neural tube defect, Down syndrome, and OFC (108, 109). Services and treatment for children with OFC can vary depending on the cleft severity, presence of associated syndromes and/or other birth defects, and the child's age and needs. For example, a child who has CL only may not need as many services compared to a child with multiple anomalies (8). However, some general recommendations exist for services and treatment for children with craniofacial anomalies such as OFC (8, 109-111). These recommendations were originally set forth by the ACPA in 1993 and were amended in 2000 and late 2004 (109). Initial evaluations of infants with OFC are recommended within the first few days of life and subsequent evaluations should be scheduled at regular intervals. The frequency of evaluations is contingent on the cleft severity and child's age.

At the first visit, a full pediatric evaluation, including nutritional and feeding assessments and a medical history, should be conducted. Genetic / dysmorphology screening and subsequent referrals for complete genetic evaluations should also be conducted at the initial visit. Infants and children with OFC need genetic services to determine the presence of associated syndromes, complexity of care involved, and the family and child's needs such as care coordination or further treatment (8). Undetected associated syndromes or other birth defects can increase the risk of developmental disabilities, speech disorders, and airway compromise (8).

Other evaluations that should occur during the initial visit include otolaryngologic, audiologic, prelinguistic speech-language, psychosocial, dental, and surgical. Audiologic and otolaryngologic services are recommended because of increased risk of hearing loss during infancy for children with OFC (8). Dental services are recommended due to the increased risk of dental caries and other potential dental problems such as missing, malpositioned or malformed teeth (11, 112-114). Well-child care or preventive services are also recommended to help establish a community medical home. The medical home is a concept developed by the American Academy of Pediatrics for all children that includes care that is accessible, continuous, coordinated, family-centered, comprehensive, culturally effective, and compassionate (30, 115-123).

In the 2000 ACPA recommendations, all of these services are recommended within the first year of life, except primary cleft surgery and otolaryngological and genetic services. Otolaryngological services are recommended within the first six months of life. Surgical closure of the CL should occur within six months of life and closure of the palate should occur within 18 months of life (109). The timeliness of cleft surgical repair is essential to

improve speech, language, dental, and psychosocial outcomes. For example, children who have their palate closed before the age of one usually develop normal speech earlier and easier than children who have their palate closed after that age (9). Per the 2000 ACPA guidelines, genetic services should occur within the first two years of life (109). Table 1.1 lists an abbreviated version of the 2000 ACPA recommended services, treatment and frequency of these services for children with OFC (109).

Table 1.1. A summary of the 2000 American Cleft Palate-Craniofacial Association recommended services and treatment for children with orofacial clefts

Type of Service	Timing / Frequency
Primary cleft/lip surgical repair	Within 18 months of life
Audiological assessment	First assessment within first year of life and at least once per year thereafter
Dental care, including primary care, routine maintenance, and orthodontic care	At least once per year throughout lifetime
Speech and language pathology, including laryngeal function	At least once per year until age four
Genetic screening, including follow-up evaluations	Within first two years of life and until puberty for some children
Otolaryngologic care	Within first six months of life and continuously through adolescence
Nursing and pediatric care	Continuously throughout life
Psychological and social services, including screening evaluations	Periodic through adolescence

As demonstrated, the ACPA recommends services and treatment throughout childhood and adulthood for children with OFC and their families. These services and treatments include surgery, audiologic, nursing, otolaryngologic, dental, genetic / dysmorphologic services, speech-language pathology, and psychological and social services.

The 2000 ACPA recommendations were utilized in this dissertation research because they were most relevant to children with OFC born between 1995 and 2002, which was the study

period. A few modifications to these recommendations were made in late 2004. The revisions applicable to this dissertation research were primary CL surgery within the first year of life instead of the first six months of life and initial audiological assessment in the first three months of life instead of within the first year of life. To date, no study has examined the timeliness of such services in accordance with the ACPA guidelines. This dissertation is therefore the first to assess timeliness of services among children with OFC.

Treatment of children with OFC has traditionally relied upon craniofacial teams or craniofacial centers. Craniofacial centers provide a coordinated, interdisciplinary team approach to care for families of children with craniofacial anomalies. The interdisciplinary teams are comprised of physicians and health care professionals from different specialties, such as audiology, genetics, neurosurgery, oral and maxillofacial surgery, orthodontics, and social work (6, 124-126). To date, no study has examined how distance to craniofacial centers or teams may affect timely receipt of services among this population. This dissertation is the first to assess how time traveled might affect timely receipt of primary cleft surgery for children with OFC, which is a gap in the literature.

Referral to Services among Children with Orofacial Clefts

One factor that can affect receipt of services, thereby affecting health service utilization and cost among children with OFC, is referral to services. To date, only four studies have examined referral to services among children with birth defects, two of which focus on children with OFC (32, 127-129). The studies on children with OFC were conducted in Maryland in the late 1960's and in Florida in the mid 1990's.

Despite being outdated, the study conducted in Maryland was the first to examine referral rates to services (32). This study examined individual and system characteristics of children with OFC and referral to the Maryland Crippled Children's Service Program. Individual characteristics consisted of cleft type, presence of other anomalies, age referred to services, and birth weight. System characteristics included source of prenatal care (PNC), initiation of PNC, and distance traveled to treatment. In general, children diagnosed with CLP utilized the Maryland Crippled Children's Service Program more compared to children diagnosed with CL or CP only (32). Of children with OFC, only 36% were known to the Maryland Crippled Children's Service Program. Only 2.3% of children received their first Maryland Crippled Children's Service Program visit by the age of 12 months. Furthermore, only 21.7% were enrolled in the Maryland Crippled Children's Service Program by the end of their fifth birthday, and 15.9% of the children received their first Maryland Crippled Children's Service Program visit between the ages of 16 and 20. In this study, the age at identification by the CCSP was significantly associated with residential area, cleft type, presence of other anomalies, and presence of other anomalies in the family (32). The authors also found that even though 34.9% of children resided within the immediate area of the facility from which treatment and services were provided, 23.3% traveled a mean one-way distance greater than 20 miles to receive services and treatment for their child (32).

Results from the study conducted in Maryland indicate that children with OFC were not receiving services and treatment in a timely fashion nor in accordance with the ACPA guidelines. However, this was prior to the implementation of such guidelines and GIS, which allows for more precise estimates of distance traveled to health care providers such as craniofacial centers and teams.

Major strengths of the study conducted in Maryland included multiple-source case ascertainment using birth certificates, the Crippled Children's Service Program registry, specialty hospital records, discharge diagnoses, and records from local health departments. This improved data accuracy and led to more accurate prevalence rates of children with OFC. Selected individual characteristics such as birth weight, cleft type and condition, entry age to the Maryland Crippled Children's Service Program, and system characteristics like source of PNC were also examined. More importantly, hospitalization rates and mean distance traveled to receive treatment were examined in this study. To date, this is the only study that has examined distances to health care providers and facilities among children with birth defects.

Despite these strengths, the study conducted in Maryland only analyzed financing and service utilization variables for 56 randomly selected cases. In addition, discussion of methodological aspects of analyzing the distance to service agency was minimal and assumptions were made with respect to residential immobility during the seven-year study period. Lastly, because this study was conducted in the late 1960's, certain geographic and demographic characteristics were not controlled for in this study, and the results are outdated (32).

The other study on referral to services among children with OFC was conducted in Florida from 1996-1997 by Williams et al. The objective of this study was to determine the referral and treatment patterns of live born Florida children with OFC identified by the Florida Birth Defects Registry. Orofacial cleft diagnoses were classified using the International Classification of Diseases, 9th revision, and reviewed by a clinical geneticist. In this study, 42% of children with OFC were evaluated or known to the Children's Medical Services'

craniofacial centers or cleft palate clinics, and 26% of children with OFC had their first contact with the craniofacial centers or cleft palate clinics within the first two months of birth (129). Almost 95% of children with OFC were first seen when they were less than three years of age. Children with CLP were significantly more likely to have had contact with the centers or clinics compared to children with CL only or CP only. The craniofacial centers and cleft palate clinics were most likely to provide evaluation between the ages of two months and three years old (129).

These results were substantially different from the study conducted in Maryland where less than three percent of children less than 12 months old with OFC were evaluated. Results from the study conducted in Florida also indicated that children with OFC whose mothers were older and White were not receiving needed services and treatment. More importantly, these data demonstrate that maternal age and race and cleft type and severity can influence referral to services and thus service use and health care costs. Additionally, these results provide rationale to include these covariates when examining health service utilization, costs, and receipt of services among this population.

Despite the strengths of the Florida study conducted by Williams et al., the authors were unable to obtain maternal socio-economic information, which could explain the phenomenon of mothers less than 25 years old being more likely to be contacted by the Children's Medical Services' (129). In addition, other potential confounding factors were not controlled for in these analyses. Such factors included marital status, number of living children, maternal education, distance to provider, and previous use of health care services like Special Supplemental Nutrition Program for Women, Infants, and Children (WIC), PNC, and MCC.

All these factors are important considerations for referral, access, utilization, and cost of health services.

Potential Barriers to Care among Children with Birth Defects

Many financial and non-financial factors can impede health service utilization and receipt of timely services among children with OFC. Financial barriers to care are well documented for CSHCN. Financial barriers to care include lack of health insurance or, as in this dissertation, low or no reimbursement from Medicaid for specialized services and treatment such as dental and orthodontic care (126, 130-132). Non-financial barriers include structural and personal barriers (98, 133-136).

Structural or system barriers stem from health policies, characteristics of service delivery, structure of the delivery system, organization of services, availability of providers and multidisciplinary teams to treat children with OFC, and availability of referral systems to identify and refer children with OFC to craniofacial centers and other specialized services (85, 98, 133-137). Such barriers result in children with OFC being misidentified, unidentified, or identified later in the developmental stages. Improper identification can be due to cost, the wrong tools being employed to identify high-risk populations, or lack of coordinated care (138). Other structural or system barriers include communication and awareness of coordinated care for related services in health and special education. Family support has also been identified as a barrier to implementing a medical home (115, 116). The location of craniofacial teams and centers in certain geographic locations of the state may also be a structural barrier (117). Despite the *Healthy People 2010* objectives 21-15 and 21-16 which promote efforts to increase the number of states that have a referral system in place to refer children with OFC to craniofacial rehabilitative teams and to conduct craniofacial

health surveillance, only 23 states and the District of Columbia had such systems in place in 1997 (139). Hence, the lack of referral systems for children with OFC may also be a structural barrier.

Personal barriers can entail parents of children with OFC not knowing their insurance will pay for certain services or the inability to navigate the health care system and use health care services effectively (98, 133-136). Other personal barriers can include transportation, appointment timing, missed work and school days, service location, and parental demographics such as low socioeconomic status. Parental and child attitudes and beliefs like distrust among health care professionals, family dynamics, and cultural factors such as language and cultural norms are also personal barriers that could contribute to decreased access to care for children with OFC.(98, 133-136).

While both financial and non-financial barriers can lead to decreased access to services among CSHCN, including children with OFC, these barriers were unable to be examined in this dissertation due to unavailability of the data. However, they are worth noting due to their likely influence on the results presented in Chapters Two and Three.

Significance of Study

Identifying trends in specific health service use and costs among different age groups of children with OFC will provide much needed information on this population, particularly in North Carolina. To date, no such data exist on health service utilization and costs by service category among this population compared to children without OFC. This dissertation examines differences in health service use and cost by cleft type and the presence of other anomalies, which is not currently addressed in the literature. These data are useful in

determining which cleft types contribute most to health service use and cost. This is also important when developing programs such as care coordination and targeting resources for children with multiple conditions. This research will provide important information on maternal, child, and system characteristics and their effect on different categories of health care service utilization and Medicaid costs. By determining which populations are underutilizing services and if any geographic variations exist in service provisions, resources can be targeted to populations in need of essential services. This dissertation also provides an overview of the utilization and financial burden in the early years of life for children with OFC compared to unaffected children. In addition, determining factors that mediate Medicaid costs can guide future investments in programs that coordinate care or otherwise serve families of children affected by OFC. Furthermore, better estimates of Medicaid costs for children with birth defects such as OFC are important for developing policies and programs in North Carolina as well as across the United States. Determining the proportion of children that received treatment and services within the ACPA guidelines and factors associated with timely receipt of services among children with OFC will provide important information for service planning, program planning, and policy development. This information is currently not addressed in the literature. Information from this dissertation should ultimately improve access to services and the overall health and development of children with OFC in North Carolina.

Conceptual / Theoretical Model

The theory that informed this dissertation was Andersen's Behavioral Model of Health Service Utilization. The Behavioral Model of Health Service Utilization was originally

developed over 35 years ago, but has been modified several times and still maintains its relevance today (140, 141). This model is commonly used to describe, predict, and explain population-based health care service utilization and medical care.

The Behavioral Model of Health Service Utilization posits that use of health services is a function of individuals' predisposition to use services, factors which impede or enable use, and need for care (140-143). Accordingly, the three components that comprise the Behavioral Model of Service Utilization are predisposing, enabling, and need characteristics (141). Predisposing characteristics are a function of individuals' predisposition to use services and include basic demographic factors like age and gender; social structural variables such as education, race/ethnicity, and marital status; and beliefs like general beliefs, values, and attitudes about the value of health services and/or availability of care (140-143). Factors such as marital status, race/ethnicity, and education imply the importance of life style and environmental influences on individuals' decisions to seek care (142). Enabling resources affect access and use of services, such as personal, family, and community factors. Community factors can include attributes of the region or community where an individual resides. For instance, in this dissertation, perinatal care region and place of residence may indicate geographic proximity to a source of care and/or attitudes about health care (142). This component also includes community resources such as provider availability and service program participation as with MCC and PNC. It also includes factors such as health insurance, income, regular source of care, transportation, and travel times (141-143).

In the Behavioral Model of Health Service Utilization, the need factor tends to be the single most important predictor of health service utilization and can be based on self-perception of one's health, clinical diagnoses and/or evaluations by health care professionals

(142). The need component includes two categories, which are perceived and evaluated need. Perceived need is largely a social phenomenon that when modeled correctly should be explained by social structural factors like race/ethnicity, education and health beliefs (141). Perceived need also includes the extent to which parents worry about their child's health, particular symptoms or conditions experienced as a result of their child's condition, and limitations on daily activities imposed by their child's condition (142, 143). Perceived need contributes to understanding the care-seeking process and adherence to medical treatment (141). In this dissertation, variables categorized as a perceived need include initiation of PNC in the first trimester, receipt of MCC, and service type. Evaluated need includes professional judgment and objective measurement about a patient's physical status and medical care. It also is related to the severity of the condition and thus the type and cost of treatment received (141, 143). In this dissertation, the presence of other anomalies (isolated versus multiple anomalies), cleft type (CL, CP, and CLP), and service type represent evaluated need. In previous studies, enabling and need factors have explained more of the variation in health service use than predisposing characteristics (142). Predisposing, enabling, and need factors will differ in their ability to explain health service utilization and expenditures depending on what service type and expenditures are being analyzed. For example, medical services utilized due to more serious and complex conditions such as CLP with another birth defect could be primarily explained by demographic and need characteristics. In contrast, dental services may be better explained by social structure, enabling resources, and health beliefs (141).

The Behavioral Model of Health Service Utilization also includes external environmental factors. These factors affect the health status of individuals within the community. They

include health care system characteristics such as policies, organization, resources, and financial arrangements, which influence the availability, accessibility, and acceptability of health care services (141). External environmental factors will ultimately influence personal characteristics such as predisposing, enabling, and need factors. This dissertation did not examine external environmental factors, but such factors may modify the effect of personal characteristics on health service utilization and thereby expenditures.

Predisposing, enabling, and need factors have been assessed in previous studies on health service utilization and costs of CSHCN. In a study conducted by Aday et al. in 1993, predisposing factors included child's age, gender, race/ethnicity, family size, family structure, and maternal education. Enabling characteristics included overall health status, perceived health, limitation(s) in major activity, and number of chronic conditions. Need factors consisted of poverty level and parental employment status (88). The authors found that number of chronic conditions, race/ethnicity, maternal education, and child's age were significant predictors of hospitalizations, use of prescribed medication, and seeing physicians in the past year (88).

In 2002, Newacheck et al. also examined predisposing, enabling, and need factors with regards to health service utilization among CSHCN. In this study, predisposing factors were family structure, child's age, sex, race/ethnicity, and education of the family reference individual. Enabling factors were family income, region of residence, place of residence, and health insurance status. Need factors were bed days due to illness and perceived health status. In this study, both the enabling and need factors differed from the factors in the 1993 Aday et al. study. Newacheck et al. found minorities were significantly more likely than White children to be uninsured (13.2% and 10.3% respectively) and without a usual source of care

(6.7% and 4.3% respectively). They also found minority children were more likely than White children to report inabilities to obtain needed medical care (3.9% and 2.8% respectively). In addition, White CSHCN were significantly more likely than minority CSHCN to have used physician services (88.6% and 85.0% respectively). After adjusting for predisposing, enabling, and need factors, racial and ethnic differences were attenuated, but remained significant for several variables such as without usual source of care, no regular clinician, and volume of physician contacts (83).

In a study conducted by Weller et al. in 2003, predisposing, enabling and need factors were assessed to determine utilization of medical and health-related services among school-aged children and adolescents with special health care needs. Similar to the studies by Newacheck et al. and Aday et al., the predisposing factors were child's age, sex, race, parental education, family size and structure. Enabling characteristics were poverty status and type of health insurance. Again, similar to the previous studies, need factors included activity limitations and perceived health status. Weller et al. found child's age, parental education, race/ethnicity, and type of health insurance were significant predictors in health service utilization among this population. However, need factors were the strongest predictors of health service use (26).

In 2004, Mayer et al. also examined predisposing, enabling, and need factors among CSHCN with respect to unmet need for routine and specialty care. Predisposing characteristics included gender, race/ethnicity, and maternal education. Enabling factors were usual source of care setting, family poverty status, and insurance status. Need factors included stability and severity of the child's condition, which differed from previous studies. Unlike in the previous studies, child's age was also considered a need factor. Mayer et al.

found that, in general, race/ethnicity, maternal education, poverty status, and severity of the child's condition were significant predictors of health service utilization (28).

In 2005, Kane et al. examined predisposing, enabling, and need factors associated with health care access for CSHCN in Mississippi. In this study, predisposing factors included the mother's education level and the child's age, sex, race, and residence. Need factors consisted of the parent's perception of the severity of the illness and illness stability. Enabling factors were usual source of care, out-of-pocket costs, income, and insurance. The authors found discontinuous insurance coverage was a significant factor associated with not having obtained routine care, and children with a high illness severity rank were more likely not to have obtained routine care than children with a low illness severity rank (100).

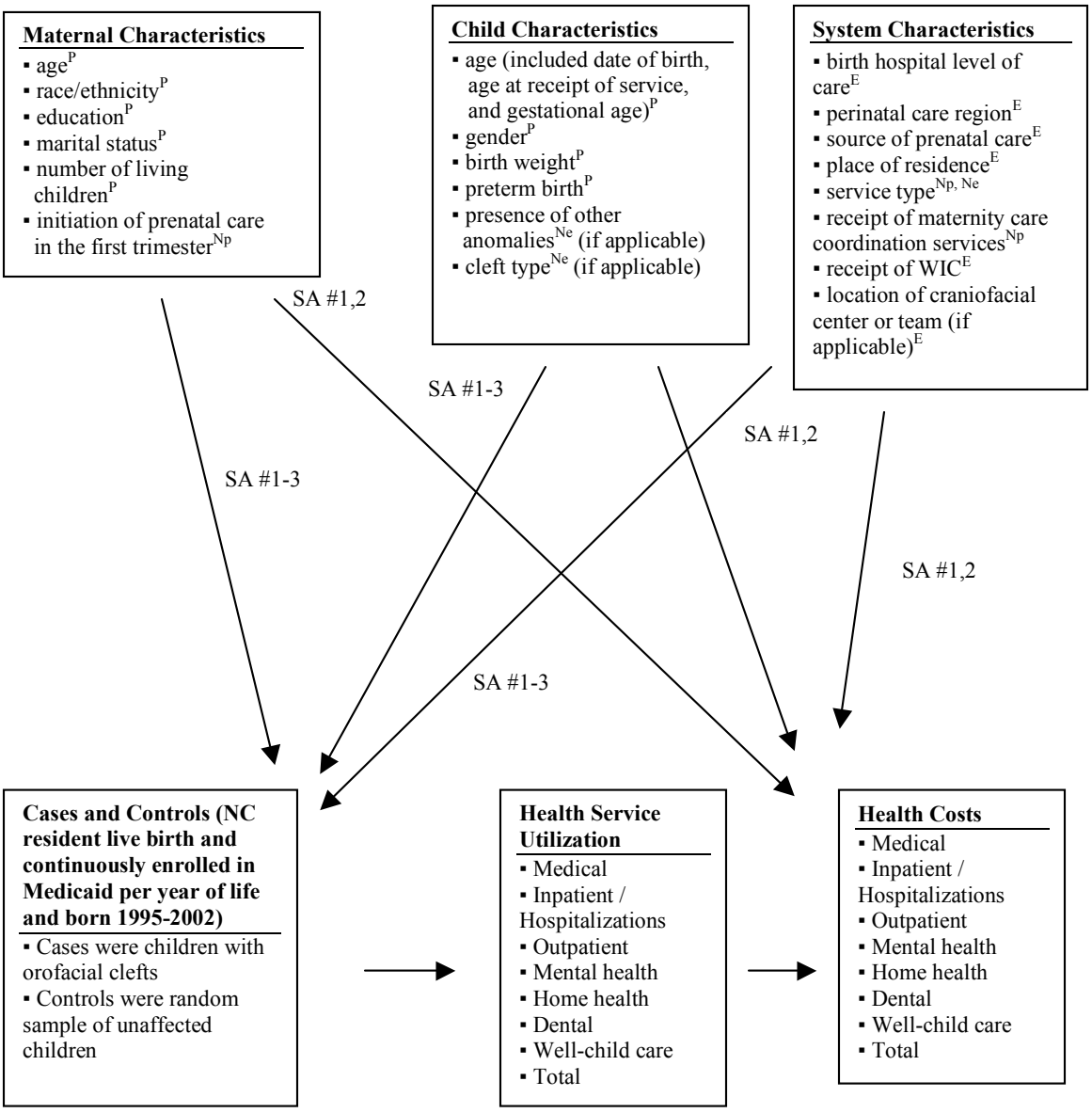
The results of these studies are congruent with previous studies that have demonstrated sociodemographic (e.g., child's age and parental education) and health-related need (e.g., severity of condition) factors are significant predictors of health service utilization and cost. As a result, the Behavioral Model of Health Service Utilization was used in this dissertation as portrayed in Figure 1.1.

The conceptual model in Figure 1.1 addresses specific aims 1-3 and defines maternal, child, and system characteristics. These aims address health service utilization and costs among cases and controls and timely receipt of services in accordance with the 2000 ACPA guidelines for children with OFC. In this dissertation, it was hypothesized that previous health service use would increase health care service utilization and thereby costs. In addition, it was hypothesized that younger mothers, less educated mothers, mothers of minority race, and children with CLP and another birth defect would utilize services more and have higher costs than their counterparts. Variation in service use among the different

perinatal care regions and places of residence due to health service availability or unavailability such as lack of craniofacial centers in some of these areas was also hypothesized. Birth hospital level of care, a system characteristic and enabling factor, was also suspected to influence health service use and cost. For example, children born in tertiary care centers would have higher service use and costs than children born in community hospitals.

Information on all of the variables presented in Figure 1.1 was available for bivariate and multivariate analysis. Medicaid status and maternal education are sometimes used as proxies for socioeconomic status. Because this analysis primarily focused on children continuously enrolled in Medicaid, Medicaid status was not assessed as a potential confounder in the analysis. The categorization used for each of these variables is discussed in the Overview of Methods section.

Figure 1.1. Conceptual model



Legend
SA = specific aim
P = predisposing characteristic
E = enabling factor
Np = perceived need
Ne = evaluated need

Overview of Methods

Research Design

Study Design

The primary goal of this dissertation is to examine patterns and predictors of health service utilization and costs among children with and without OFC on Medicaid. This goal is addressed in Chapter Two. A secondary goal is to examine the timeliness of services among children with OFC in accordance with nationally recommended treatment guidelines, which is discussed in Chapter Three. The study design of Chapter Two is a retrospective, case-control study of children born in North Carolina during January 1, 1995, through December 31, 2002. For Chapter Three, the study design is a retrospective, cohort study of children with OFC born during the same time period.

Sample Selection

Mothers eligible delivered a live infant in North Carolina during the study period. The unit of analysis was the woman or child depending on the variable examined, but in most cases, it was the child. Cases were defined as children with OFC born during this time period and were ascertained by the North Carolina Birth Defects Monitoring Program (NCBDMP) using British Pediatric Association codes 749.000-749.290. The date of OFC diagnosis was defined as the infant's date of birth verified by medical record and/or hospital discharge records because OFC are readily apparent at birth.

The control sample consisted of children who were live born during the same time period without OFC and/or another birth defect. These children were randomly matched in a 10:1 ratio by birth cohort to cases to increase power. This ratio was chosen to ensure sufficient

numbers for any effect measure modification or subgroup analysis. Because the probability of inclusion of children with OFC or children with OFC and another birth defect was less than three percent in the control group, these children were not excluded upfront from the random selection of controls.

Other inclusion requirements for both cases and controls included children continuously enrolled per year of life in Medicaid through age four. The restriction to children on Medicaid was to control for low socioeconomic status, as all children on Medicaid are families of relatively low income. Children's use of health care service varies substantially by type of health insurance coverage (78). Thus, by restricting the study sample to only one type of health insurance, the independent effects of health insurance coverage were controlled. In addition, it was the only database available to track services for this population. Exclusion criteria for cases and controls included infant death within 12 months of life, out-of-state delivery and/or resident, and any adopted children.

For this dissertation, only children continuously enrolled in each year of life in Medicaid were analyzed to have a more complete depiction of service use and cost. Continuous enrollment in Medicaid was defined as enrollment 11 or more months in any given year of life. This is a commonly used definition for continuous enrollment in Medicaid and utilized as the standard definition for the Health Plan Employer Data and Information Set, which is the most widely used set of performance measures in the managed care industry (66, 144, 145). Medicaid enrollment records were obtained for January 1, 1995, through December 31, 2004, which allowed for examination of at least two years after birth of Medicaid enrollment for every child. For each child during this time period, a monthly Medicaid enrollment history indicating Medicaid enrollment (yes/no) was created.

For 1995-2002 in North Carolina, there were 1,355 children with OFC. Of these children, 103 died during the first year of life and thus were excluded from the analysis. Of the remaining 1,252 children with OFC for 1995-2002, 50.1% of children with OFC were on Medicaid (n=627).

The original control sample was 6,270 children on Medicaid. After excluding controls that had a diagnosis of OFC, other major birth defects, or died during the first year of life, the control sample size was 6,127 children.

During the first year of life, 90.1% of children with OFC (n=565) and 92.6% of unaffected children (n=5,674) were continuously enrolled in Medicaid. Enrollment in Medicaid for the cases decreased by 25.0% during the second year of life (65.1% were enrolled) and continued to decrease by about 9.0% per year of life. Similar patterns of decreased continuous enrollment in Medicaid were observed among the controls through age four. Continuous enrollment for controls decreased 32.5% in the second year of life and continued to decrease by about 8.0% each year of life through age four. The numbers of continuously enrolled children and selected maternal, child, and system characteristics per year of life are in Appendix D. These Medicaid enrollment patterns were congruent with previous studies on patterns of Medicaid enrollment (146).

Description of Data Sources and Matching Procedures

Data sources used in this dissertation included the North Carolina BabyLove files, the NCBDMP, and Medicaid enrollment records and Medicaid paid claims. The BabyLove files are composite North Carolina birth files matched with Medicaid newborn hospitalization records, maternal delivery records, and maternity case management records. These files

contain information on vital statistics such as infant's date of birth, maternal age, maternal race/ethnicity, maternal education, receipt of PNC, and other demographics from the birth certificate and fetal death certificate. The BabyLove files also include data on child service coordination, WIC, Medicaid status, PNC, MCC services, Medicaid newborn costs in the first 60 days of life, and Medicaid infant costs in the first year of life.

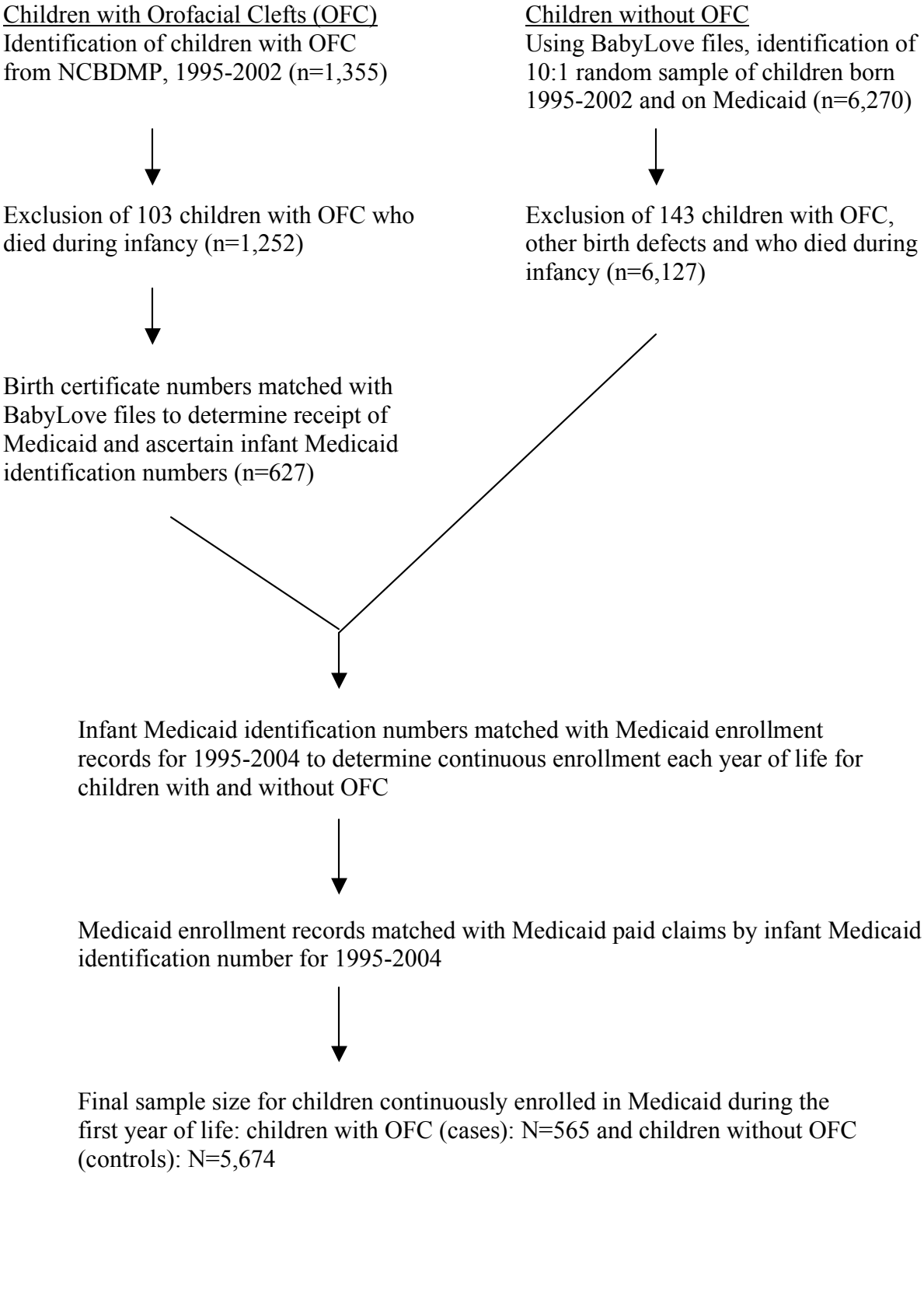
The NCBDMP is located at the State Center for Health Statistics within the North Carolina Division of Public Health. The program was started in 1987 and is a population-based, statewide program that utilizes a combination of passive and active ascertainment. Passive ascertainment entails the linking with other administrative data sources such as vital statistics, BabyLove files, and hospital discharge data. Active ascertainment includes trained personnel reviewing and abstracting data from all North Carolina hospitals that provide labor and delivery and pediatric services. The program covers about 100 hospitals, excluding military hospitals, and approximately 120,000 resident births per year. The program includes all resident live births, fetal deaths of 20 or more weeks gestation, and therapeutic abortions at any gestational age. Most major birth defects are ascertained, including over 200 types of structural defects. Infants are ascertained up to one year after delivery (42).

Medicaid enrollment records and paid claims comprised the remaining data sources. These two Medicaid files are relational databases that originate from the North Carolina Division of Medical Assistance, but are housed on the State Center for Health Statistics mainframe. Information in the Medicaid enrollment records included enrollment dates, Social Security Income status, provider counties, eligibility history dates, and special needs. Medicaid enrollment records were used to determine continuous enrollment in Medicaid between 1995 and 2004. Medicaid paid claims included information on claim type, service

dates, diagnoses, procedural and provider specialty codes, discharge information, and costs. Health care service utilization and costs by service category were tracked using Medicaid paid claims. Records of all services received and paid for by Medicaid for each child with OFC and for each unaffected child born between 1995 and 2002 were extracted. To allow for at least two years of health care utilization and costs for all children with OFC (cases) and a random selection of children (controls) born in this time period, Medicaid paid claims January 1, 1995, through December 31, 2004, were employed.

Figure 1.2 describes the matching procedures that were utilized for this dissertation research. First, children with OFC who were born between 1995 and 2002 were identified by the NCBDMP using the British Pediatric Association coding for OFC (codes 749.000-749.290). Then, the cases were matched to the BabyLove files using the birth certificate number to determine infants with OFC on Medicaid and infant Medicaid identification numbers, which yielded a 100% matching rate. To determine the control sample, a random sample of infants on Medicaid born 1995-2002 were drawn from the BabyLove files along with infant Medicaid identification numbers. Next, the BabyLove files were matched to the Medicaid enrollment records using the infant Medicaid identification number. The matching rate was 98.5%. Finally, Medicaid enrollment records were matched to Medicaid paid claims using the infant Medicaid identification numbers. The matching rate was 100.0%.

Figure 1.2. Matching procedures



Description of Variables Analyzed

Assessment of Primary Outcomes (Dependent Variables)

To address specific aims one and two in Chapter Two, the primary outcomes of interest were medical, inpatient/hospitalizations, outpatient, mental health, home health, dental, well-child, other, and total service utilization and cost. These categories were analyzed dichotomously and continuously. These categories have been employed in previous studies on health service utilization and expenditures (21, 28, 66, 77, 95, 145). Health care service utilization and costs were analyzed for each year of life for each child. Data elements for health service utilization and costs were obtained from the Medicaid paid claims records. Medicaid paid claims types were already categorized into medical, inpatient, outpatient, dental, well-child care, outpatient, home health, and other. Medical paid claims were defined as any professional claims or fees associated with office based physicians' appointment(s) as well as professional claims associated with any outpatient and inpatient visit. Inpatient paid claims included any hospital and facility fees such as room use, labs, and X-rays associated with an inpatient visit. Outpatient paid claims consisted of any hospital and facility fees associated with an outpatient visit. Outpatient claims also included any emergency department claims. "Other" claim types included professional crossover, medical vendor, and outpatient and inpatient crossover paid claims. Total paid claims consisted of the sum of paid claims of all the service categories, and total costs included the sum of all the costs of all the cost categories. Because mental health claim types do not exist, this category was created using service categories, provider specialty codes (also known as billing provider specialty codes), provider type codes, diagnostic related group codes, and procedural codes that referred to mental retardation, developmental disabilities, and substance abuse services. The

creation of the mental health variable was in accordance with guidelines from the North Carolina Division of Mental Health**,** Developmental Disabilities and Substance Abuse Services. For purposes of this research, the number of paid claims in each service category was a proxy for health service utilization in that category.

To address specific aim three in Chapter Three, the primary outcomes of interest were timely receipt of primary cleft surgery, otolaryngologic care, audiological services, speech and language therapy, dental services, genetic services, and psychological and social services. To create these outcomes using the Medicaid paid claims, all past and current procedural terminology and diagnostic-related group codes for these services were employed. For dental services, the Medicaid paid claim pre-established category for dental claims and all past and current procedural terminology and diagnostic-related group codes related to dental and orthodontic care were used. To ensure a comprehensive inclusion of procedural codes used during the study period, consultations occurred with several members of craniofacial centers and teams in the state. Any procedural codes for these services used by these craniofacial centers and teams and the Children's Developmental Services Agency for reimbursement of Medicaid were also included. Children's Developmental Services Agencies implement the Infant-Toddler Program in each region of North Carolina, which is the state's interagency system of early intervention services for children aged birth to five years old with special health care needs. Four procedural codes commonly used for generic office visits, 99202, 99212, 99213, and 99214, were excluded unless the specific service rendered could be determined from the provider specialty code.

Assessment of Selected Maternal, Child, and System Characteristics (Independent Variables)

All of the information on selected maternal, child, and system characteristics came from or were created from variables found in the BabyLove files or the NCBDMP. The variable for time traveled to a craniofacial center or team, which was used in the GIS analysis, to assess receipt of timely cleft surgery was the exception. All of these characteristics were used in both Chapters Two and Three except the time traveled variable, which was only used in Chapter Three because geocoding was unavailable for the children without OFC.

Some sociodemographic information from the birth certificate such as maternal age, race/ethnicity, education, and marital status were collected based on maternal self-report. Other covariates such as birth defect diagnoses and presence of other anomalies were collected from the NCBDMP data. A more detailed definition of the selected maternal, child, and system characteristics and description of the data source, original coding, and coding used in this dissertation are discussed below and summarized in Table 1.2. Covariates were categorized into individual (maternal and child) and system characteristics and were defined as follows:

- *Maternal characteristics:* age, education, race/ethnicity, number of living children, marital status, and initiation of PNC in the first trimester
- *Child characteristics*: age (including gestational age and age at receipt of services), birth weight, preterm birth, gender, and for children with OFC, cleft type and presence of other birth defects
- *System characteristics*: service type, PNC source, receipt of MCC services, birth hospital level of care, perinatal care region, place of residence, receipt of WIC, and for children with OFC, time traveled to a craniofacial center or team

Age: Both the mother's age and child's age were obtained from the BabyLove files. The mother's age was first coded as a continuous variable and then as a categorical measure.

These categories were less than or equal to 20, 21-24, 25-29, and greater than or equal to 30 years old. The child's age, including gestational age in weeks, was first coded as continuous and then as a categorical measure. The child's age was assessed as the following: 0-11 months, which was the first year of life; 1-year-old (12-23 months of age), which was the second year of life; 2-years-old (24-35 months of age), which was the third year of life; 3-years-old (36-47 months of age), which was the fourth year of life; and 4-years-old (48-59 months of age), which was the fifth year of life. This was based on the child's date of birth and year of life when continuously enrolled in Medicaid. These categories of age are based on previous studies on health service utilization and costs (21, 66, 77, 145). Different age groups were examined because of potential differences in health service use and cost and because differences exist between age groups with the recommended ACPA treatment for children with OFC.

Maternal education: This covariate came from the BabyLove files as a continuous variable as years of education. Then, it was categorized into less than high school (less than 12 years of education), high school (12 years of education), and greater than high school (greater than 12 years of education).

Maternal race/ethnicity: From information in the BabyLove files, this covariate was created from the race and Hispanic origin variable and coded as White/Non-Hispanic, Black/Non-Hispanic, Hispanic, and Other. The "other" category included Native American, Asian/Pacific Islander and other non-White and had relatively few records. This variable was coded as a nominal categorical variable.

Marital status: This variable was obtained from the BabyLove files and was coded as a nominal categorical variable, married or not married.

Number of living children: This variable was obtained from the BabyLove files as a continuous variable and was coded as an ordinal categorical variable as zero, one, or greater than or equal to two.

Prenatal care: The BabyLove files included information on the month PNC began and source of PNC for each mother. The month PNC began was originally a continuous variable and was used to create the initiation of PNC in the first trimester variable. Source of PNC was analyzed as a dichotomous variable as health department or other.

Infant birth weight: This covariate was obtained from the BabyLove files and initially was assessed as continuous and then recoded as an ordinal category as less than 2,500 grams, which was born low birth weight (LBW) and greater than or equal to 2,500 grams, which was not born LBW.

Preterm birth: From the BabyLove files, gestational age in weeks was obtained to create the preterm birth variable. Preterm birth was defined as less than or equal to 36 weeks and was analyzed as a dichotomous variable.

Cleft type: For children with OFC, this variable came from the NCBDMP data. The cleft types were defined using the British Pediatric Association codes 749.100-749.190 for CL, 749.000-749.090 for CP, and 749.200-749.290 for CLP.

Presence of other anomalies: This variable was created from the NCBMDP data into isolated anomaly and multiple anomalies. Isolated anomaly was defined as children with OFC only. Multiple anomalies were defined as a child with OFC and one or more birth defect using the British Pediatric Association codes for reportable birth defects in North Carolina.

Maternity care coordination (MCC) services: A cornerstone of the Baby Love Program is MCC services that address medical, nutritional, psychosocial, and resource needs such as

payment for checkups and assistance with transportation, childbirth and parenting classes, hospital care for the infant's delivery, health care for the woman and infant after the infant's birth, and referral to other programs through the first 60 days of the infant's life (147). The Baby Love Program is one several programs in North Carolina that identify high-risk women and children for services. Receipt of MCC services was collected from the BabyLove files and was analyzed dichotomously in the analyses.

Birth hospital level of care: From data in the BabyLove files, this variable was created using the county of occurrence, hospital, and bed codes. It was recoded as a nominal categorical variable as tertiary care centers or community / other hospitals.

Perinatal care region: These regions were established in the 1980's in an effort to develop regional referral networks of perinatal care in North Carolina (148, 149). Created from the county of occurrence variable in the BabyLove files, these regions were analyzed as a nominal categorical variable as northwestern, northeastern, southeastern, southwestern, eastern, and western. Appendix G indicates which counties are included in each perinatal care region.

Place of residence: From the BabyLove files, the maternal county of residence was used to create a modified version of the Urban Influence Codes. These codes are based on counties and classified into 12 levels that build on the Office of Management and Budget metropolitan and nonmetropolitan dichotomy (150-154). Based on the 2000 Census, the Urban Influence Codes were revised in 2003. This revised version was more representative of the study sample than using the Urban Influence Codes based on the 1990 Census. In this dissertation, the 12 levels were collapsed into four categories based on metropolitan, nonmetropolitan, adjacency, and non-adjacency areas. Adjacency includes physical adjacency and at least 2%

of the population commuting to the metropolitan or micropolitan area (154). Table 1.3 describes the original Urban Influence Coding as defined by the Office of Management and Budget. Due to small numbers, the categories were collapsed into four categories as indicated in Table 1.3.

Time traveled to craniofacial centers or teams: In North Carolina, there are seven craniofacial centers and teams. Two craniofacial centers are located in Chapel Hill and Winston-Salem. Craniofacial teams are located in Durham, Winston-Salem, Charlotte, and Greenville. Appendix H shows the locations of these craniofacial centers and teams in the perinatal care regions of the state. To determine time traveled to the closest craniofacial center or team, the maternal birth address was used from the BabyLove files in the geocoding and GIS analysis, which is described in the analysis plan for Chapter Three in the following section. This variable was categorized as traveling less than or equal to 30 minutes, 31-60 minutes, 61-89 minutes, and greater than or equal to 90 minutes. Time traveled was chosen instead of distance as it was more representative of a potential barrier to care than distance traveled.

Table 1.2. Data source, original coding, and coding used for covariates

Data Source	Original Variable	Original Coding	New Variable	New Coding
BabyLove files	Maternal age	Continuous; 0-99	Same	≤ 20; 21-24; 25-29; ≥ 30 years
BabyLove files	Maternal education	Continuous; 0-99	Same	< high school; high school; > high school
BabyLove files	Race/ethnicity	--Maternal race: White, Black, American Indian, Chinese, Japanese, Hawaiian, Filipino, other Asian, other non-White, unknown	same	White/non-Hispanic; Black/non-Hispanic; Hispanic; Other
		--Maternal Hispanic origin: Cuban, Mexican, Non-Hispanic, Other Hispanic, Puerto Rican, Central/South American, unknown		
BabyLove files	Number of living children	Continuous 00-99 and unknown	Same	0, 1, ≥ 2
BabyLove files	Marital status	Married, not married, unknown	Same	Married and not married
BabyLove files	Month PNC began	Continuous	Initiation of PNC in first trimester	1-3 months = yes; 0,4-9 and unknown = no
BabyLove files	Birth weight	Continuous	Same	< 2,500 grams = LBW; ≥ 2,500 grams not LBW
BabyLove files	Preterm birth	Continuous	Same	≤ 36 weeks = yes; > 36 weeks = no
BabyLove files	Gender	Male, female, unknown	Same	Male or female
NCBDMP	Orofacial clefts (created from birth defects data)	749.000-749.290 British Pediatric Association Codes	Cleft type	cleft lip: 749.100-749.190; cleft palate: 749.000-749.090; cleft lip with cleft palate: 749.200-749.290

Data Source	Original Variable	Original Coding	New Variable	New Coding
NCBDMP	Presence of other anomalies	Used British Pediatric Association Codes for birth defects	Same	If 749.000-749.290 only diagnosis = isolated anomaly; if have this code and other birth defects code = multiple anomalies
BabyLove files	Health services information system identification number	Absent or present	Source of PNC	If present, then received PNC at a health department; otherwise, received somewhere else
BabyLove files	Maternity care coordination identification number	Absent or present	Receipt of maternity care coordination	If present, then received maternity care coordination services; otherwise, no receipt of such services
BabyLove files	WIC identification number	Absent or present	Receipt of WIC	If present, then received WIC; otherwise, no receipt of WIC
BabyLove files	County of occurrence and hospital and bed code	1-100 codes for each county in North Carolina; hospital bed code-home or non-institutional, general hospital, TB hospital, mental hospital, chronic hospital, penal hospital, veterans hospital, nursing and rest home, clinic and doctor's office, other institution; bed code-<20 beds, 20-24 beds, 25-49 beds, 50-99 beds, ≥100 beds	Birth hospital level of care	Tertiary hospital and community hospital
BabyLove files and location of craniofacial centers and teams from ACPA 2005-2006 membership directory	Maternal residential address at birth and address of closet craniofacial center or team to maternal birth address	Address, city, state, and zip code	Time traveled	≤ 30, 31-60, 61-89, and ≥ 90 minutes (See Appendix H for location of craniofacial centers and teams in North Carolina)

Data Source	Original Variable	Original Coding	New Variable	New Coding
BabyLove files	County of occurrence	1-100 code for each county in North Carolina	Perinatal care region	northwestern, northeastern, southwestern, southeastern, western, or eastern (See Appendix G for which county is assigned to each region)
BabyLove files	County of residence	1-100 code for each county in North Carolina	Place of residence	See Table 1.3 for explanation of coding

Table 1.3. Description of 2003 Urban Influence Codes and Creation of Place of Residence Variable

Urban Influence Code	Urban Influence Codes Description	Categories for Place of Residence Variable
1	In large metropolitan area of ≥ 1 million residents	1
2	In small metropolitan area of < 1 million residents	1
3	Micropolitan adjacent to large metropolitan area	2
4	Noncore adjacent to large metropolitan	3
5	Micropolitan adjacent to small metropolitan area	2
6	Noncore adjacent to small metropolitan area with own town	3
7	Noncore adjacent to small metropolitan area with no own town	3
8	Micropolitan not adjacent to a metropolitan area	4
9	Noncore adjacent to micropolitan area with own town	3
10	Noncore adjacent to micropolitan with no own town	3
11	Noncore not adjacent to metropolitan or micropolitan with own town	4
12	Noncore not adjacent to metropolitan or micropolitan with no own town	4

Exploratory Data Analysis

Univariate distributions of each covariate were examined to identify potential outliers, note the extent of missing values, and confirm an appropriate coding scheme. In addition, univariate analyses were conducted to determine frequency distributions of services and cost, including minimum, maximum, and average per child with OFC and per unaffected child. While some covariates already were divided into categories based on the literature and well-established cut-points, others were continuous in nature. Decisions on whether to categorize any continuous variables were made based on these descriptive analyses. For binary and categorical covariates, proportions were generated using PROC FREQ in SAS. For

continuous variables with a normal distribution, the mean and standard deviation were computed and histograms and boxplots were generated using PROC UNIVARIATE in SAS. From this output, the skewness and kurtosis of the distributions were assessed.

Bivariate analyses included examining differences in the demographics of cases and controls using either chi-square or Fisher's exact test and of the various service and cost categories and maternal, child, and system characteristics among cases and controls. The bivariate distributions of the outcome by each covariate were examined to confirm final decisions on the coding of continuous and categorical variables. For instance, if bivariate analyses indicated that one level of a categorical variable had a smaller number of observations in one or both cells, a decision was made to merge categories, such as with the place of residence. The Wilcoxon rank sum test was performed to assess if the mean number of paid claims and average cost per child for children with and without OFC were identical. The Wilcoxon rank sum test is commonly applied to data that exhibit skewness and does not require the underlying distributions to be normally distributed. If cell sizes were greater than five, Pearson chi-square was employed, and if cell sizes were less than or equal to five, Fisher's exact tests were used for categorical variables to compare differences between children with and without OFC in regards to selected maternal, child and system characteristics. Pearson chi-square or Fisher's exact test were also employed to determine differences between the cleft types among children with OFC and selected maternal, child and system characteristics. For children with OFC, analyses were stratified by cleft type and by presence of other anomalies. All analyses were conducted using SAS software, version 9.1.

Aim-Specific Analyses

Aim 1: To describe health care service use and costs among children with OFC (cases) and without OFC (controls) on Medicaid during the first five years of life.

To address this aim, total use of health care services and cost and service and cost category for each year of life were assessed. The average number of paid claims and average cost per child among cases and controls were examined to determine any differences between the two groups. Total Medicaid costs for each cost category were calculated for cases and controls and for each year of life. For children with OFC, these analyses were also conducted by cleft type and presence of other anomalies. Chapter Two describes health service use and cost among cases and controls during the first year of life. Appendix E and F describe health service use and cost among cases and controls during the second through fifth year of life.

Aim 2: To determine individual and system characteristics associated with health service use and costs among children with OFC and unaffected children on Medicaid during the first year of life.

First, univariate and bivariate analyses on the primary outcomes of specific health service utilization and cost categories and maternal, child, and system characteristics were conducted as previously discussed. To examine how selected maternal, child, and system characteristics were associated with the number of paid claims (i.e., service use) for each health service category, log-linear Poisson regression was employed. Poisson regression is a form of the generalized linear model and, in this case, models the log of count ratios or number of events. Beta coefficients, standard errors, effect estimates and 95 percent CI were computed for each

covariate to assess the magnitude and precision of the effect estimates. However, only the count ratios, which are the effect estimates, and 95 percent CI are presented. All covariates were included in the model. For all the categorical variables, indicator variables were created in the model. Model equation one was used in the Poisson regression analysis for cases and controls:

Model Equation 1. $\text{Log}\,(Y_{i\,(\text{discrete count})}) = \beta_{0\,i} + \beta_1\,(\text{maternal age}_1) + \beta_2\,(\text{maternal age}_2) + \beta_3\,(\text{maternal age}_3) + \beta_4\,(\text{maternal education}_1) + \beta_5\,(\text{maternal education}_2) + \beta_6\,(\text{maternal race}_1) + \beta_7\,(\text{maternal race}_2) + \beta_8\,(\text{maternal race}_3) + \beta_9\,(\text{marital status}_1) + \beta_{10}\,(\text{number of living children}_1) + \beta_{11}\,(\text{number of living children}_2) + \beta_{12}\,(\text{initiation of PNC in 1st trimester}_1) + \beta_{13}\,(\text{birthweight}_1) + \beta_{14}\,(\text{preterm birth}_1) + \beta_{15}\,(\text{gender}_1) + \beta_{16}\,(\text{PNC source}_1) + \beta_{17}\,(\text{receipt of WIC}_1) + \beta_{18}\,(\text{receipt of MCC}_1) + \beta_{19}\,(\text{birth hospital}_1) + \beta_{20}\,(\text{perinatal care region}_1) + \beta_{21}\,(\text{perinatal care region}_2) + \beta_{22}\,(\text{perinatal care region}_3) + \beta_{23}\,(\text{perinatal care region}_4) + \beta_{24}\,(\text{perinatal care region}_5) + \beta_{25}\,(\text{place of residence}_1) + \beta_{26}\,(\text{place of residence}_2) + \beta_{27}\,(\text{place of residence}_3) + \varepsilon_i$

In this equation, the subscript i refers to the specific outcome modeled, which was medical, inpatient, outpatient, mental health, home health and total. Multivariate analyses were restricted to outcomes where there was sufficient data among cases and controls. Hence, well-child care, dental, and other health service categories were not examined in the multivariate analyses. Poisson regression models were run separately on children with and without OFC.

To determine how selected maternal, child, and system characteristics were associated with each cost category, a two-part model was employed for each outcome. A two-part model was appropriate for the cost categories because a disproportionate number of children had zero costs, which were the non-users of service. First, health care costs for each child for each outcome were created into a binary variable. Then, a binary logit model was used to examine the effect of selected characteristics on each cost category to determine the likelihood of incurring any costs by the selected covariates. Odds ratios (OR) were computed

to determine the magnitude of these relationships, and 95 percent CI were generated to examine the precision of these effect estimates. If a cost category had less than five percent of children who did not have costs, then a logit regression was not conducted, only ordinary least squares regression analysis, which is the second part of the two-part modeling. Because outpatient, mental health, and home health categories had greater than five percent of children with zero costs, these categories were the only ones examined in the binary logit analyses. All covariates were included in the model. For all the categorical variables, indicator variables were created in the model. The model equation used in the binary logit analysis is shown in model equation two.

Model Equation 2. $\text{Logit}\,(Y_{i\,(\text{dichotomous})}) = \beta_{0\,i} + \beta_1\,(\text{maternal age}_1) + \beta_2\,(\text{maternal age}_2) + \beta_3\,(\text{maternal age}_3) + \beta_4\,(\text{maternal education}_1) + \beta_5\,(\text{maternal education}_2) + \beta_6\,(\text{maternal race}_1) + \beta_7\,(\text{maternal race}_2) + \beta_8\,(\text{maternal race}_3) + \beta_9\,(\text{marital status}_1) + \beta_{10}\,(\text{number of living children}_1) + \beta_{11}\,(\text{number of living children}_2) + \beta_{12}\,(\text{initiation of PNC in 1st trimester}_1) + \beta_{13}\,(\text{birthweight}_1) + \beta_{14}\,(\text{preterm birth}_1) + \beta_{15}\,(\text{gender}_1) + \beta_{16}\,(\text{PNC source}_1) + \beta_{17}\,(\text{receipt of WIC}_1) + \beta_{18}\,(\text{receipt of MCC}_1) + \beta_{19}\,(\text{birth hospital}_1) + \beta_{20}\,(\text{perinatal care region}_1) + \beta_{21}\,(\text{perinatal care region}_2) + \beta_{22}\,(\text{perinatal care region}_3) + \beta_{23}\,(\text{perinatal care region}_4) + \beta_{24}\,(\text{perinatal care region}_5) + \beta_{25}\,(\text{place of residence}_1) + \beta_{26}\,(\text{place of residence}_2) + \beta_{27}\,(\text{place of residence}_3) + \varepsilon_i$

In this model equation, the subscript i refers to the specific outcome modeled, which was outpatient, mental health, and home health.

Binary logit models were run separately for children with OFC and unaffected children. Likelihood ratio tests were employed for each cost category to determine if each regression coefficient for maternal, child, and system characteristic in each cost category were similar for cases and controls. Likelihood ratio tests were used due to the non-normally distributed errors. Non-normally distributed errors can be a result of heteroscedasticity and skewed data, which is commonly exhibited with expenditure data.

For the second part of the two-part model, ordinary least squares regression analyses were employed conditional on those children who had costs. Using a continuous variable for each outcome, all costs were log transformed to reduce skewness. Beta coefficients, standard errors, effect estimates, and 95 percent CI were obtained to determine the magnitude and precision of the effect estimates, which were cost ratios. However, for ease of interpretation, only cost ratios and 95 percent CI are reported. The model equation used in the ordinary least squares regression analysis is shown in model equation three where the subscript i refers to the specific outcomes modeled, which were medical, inpatient, outpatient, mental health, home health and total costs.

Model Equation 3. $\text{Log}\,(Y_{i\,(\text{continuous})}) = \beta_{0\,i} + \beta_1\,(\text{maternal age}_1) + \beta_2\,(\text{maternal age}_2) + \beta_3\,(\text{maternal age}_3) + \beta_4\,(\text{maternal education}_1) + \beta_5\,(\text{maternal education}_2) + \beta_6\,(\text{maternal race}_1) + \beta_7\,(\text{maternal race}_2) + \beta_8\,(\text{maternal race}_3) + \beta_9\,(\text{marital status}_1) + \beta_{10}\,(\text{number of living children}_1) + \beta_{11}\,(\text{number of living children}_2) + \beta_{12}\,(\text{initiation of PNC in 1st trimester}_1) + \beta_{13}\,(\text{birthweight}_1) + \beta_{14}\,(\text{preterm birth}_1) + \beta_{15}\,(\text{gender}_1) + \beta_{16}\,(\text{PNC source}_1) + \beta_{17}\,(\text{receipt of WIC}_1) + \beta_{18}\,(\text{receipt of MCC}_1) + \beta_{19}\,(\text{birth hospital}_1) + \beta_{20}\,(\text{perinatal care region}_1) + \beta_{21}\,(\text{perinatal care region}_2) + \beta_{22}\,(\text{perinatal care region}_3) + \beta_{23}\,(\text{perinatal care region}_4) + \beta_{24}\,(\text{perinatal care region}_5) + \beta_{25}\,(\text{place of residence}_1) + \beta_{26}\,(\text{place of residence}_2) + \beta_{27}\,(\text{place of residence}_3) + \varepsilon_i$

Ordinary least squares regression models were run separately for cases and controls. Chow tests were employed to determine any differences between the models for cases and controls in each cost category. The Chow test is an analysis-of-variance F-ratio test, which tests for structural change in regression analysis. Specifically, it jointly tests the intercept and slope of the dummy variables. Because the Chow test assumes homoscedasticity, it was not used to compare the regression coefficients for cases and controls in the binary logit models.

Effects of individual and system characteristics on health service use and cost among cases and controls were only assessed during the first year of life, which are discussed in

Chapter Two. Due to small numbers of continuously enrolled children in both groups of children for ages one through four, these effects could not be examined.

Aim 3: To assess the timeliness of certain services according to the ACPA recommendations among children with OFC.

In Chapter Three, univariate and bivariate analyses were conducted using chi-square or Fischer's exact test to determine any differences between cleft types and selected maternal, child, and system characteristics. Effect modification by cleft type for the associations between maternal, child, and system characteristics and timely receipt of specialized services were assessed using Pearson chi-square and Fisher's exact tests using p less than 0.05 for statistical significance or Breslow-Day test of homogeneity with a cut point of p less than 0.10.

Multivariate logistic regression analysis was employed to determine maternal, child, and/or system characteristics associated with timely receipt of cleft surgery. Receipt of timely OFC surgery was categorized as a binary outcome as initial surgery occurring less than 18 months of life and initial surgery occurring at 18 months of life or later. To determine if cleft type modified the relationship between presence of other anomalies and timely cleft surgery, stratum-specific OR were examined and a cut-point of p less than 0.10 was used in the Breslow-Day test of homogeneity. Odds ratios and 95 percent CI were calculated in the multivariate logistic regression model. The model equation used in specific aim three in Chapter Three is outlined below in model equation four.

Model Equation 4. Logit (timely cleft surgery$_{(\text{dichotomous})}$)= $\beta_{0\,i}$ + β_1 (maternal age$_1$) + β_2 (maternal age$_2$) + β_3 (maternal age$_3$) + β_4 (maternal education$_1$) + β_5 (maternal education$_2$) + β_6 (maternal race$_1$) + β_7 (maternal race$_2$) + β_8 (maternal race$_3$) + β_9 (marital status$_1$) + β_{10} (number of living children$_1$) + β_{11} (number of living children$_2$) + β_{12} (initiation of PNC in 1st

$\text{trimester}_1) + \beta_{13}(\text{birthweight}_1) + \beta_{14}(\text{preterm birth}_1) + \beta_{15}(\text{gender}_1) + \beta_{16}(\text{PNC source}_1) + \beta_{17}(\text{receipt of WIC}_1) + \beta_{18}(\text{receipt of MCC}_1) + \beta_{19}(\text{birth hospital}_1) + \beta_{20}(\text{perinatal care region}_1) + \beta_{21}(\text{perinatal care region}_2) + \beta_{22}(\text{perinatal care region}_3) + \beta_{23}(\text{perinatal care region}_4) + \beta_{24}(\text{perinatal care region}_5) + \beta_{25}(\text{place of residence}_1) + \beta_{26}(\text{place of residence}_2) + \beta_{27}(\text{place of residence}_3) + \beta_{28}(\text{cleft type}_1) + \beta_{29}(\text{cleft type}_2) + \beta_{29}(\text{presence of other anomlies}_1) + \beta_{30}(\text{time traveled}_1) + \beta_{31}(\text{time traveled}_2) + \beta_{32}(\text{time traveled}_3) + \varepsilon_i$

To determine if travel time was associated with receipt of timely cleft surgery, the maternal birth residential addresses for children with OFC were geocoded, and GIS analysis was conducted. The GIS analysis was conducted using *ESRI ArcGIS* and *Network Analyst 9.2. TeleAtlas Multinet 2005 2.1* was used for the road network to match an address on a street network to obtain real-world coordinates and to be able to place that address on a map. The street network is attributed with address ranges, street names, street types, cities, and zip codes. The software then performs a "match" against the road reference and interpolates where along a road to place the point. Spatial analysis of point-to-point distance was also performed within *ArcGIS*, which provided the average distance and time traveled between residential addresses of families of children with OFC and the closest craniofacial center or team to that address.

In summary, Chapter Two examines patterns of and factors associated with different categories of health service utilization and cost among children with and without OFC who were continuously enrolled in Medicaid during the first year of life. Appendix D-F examine the demographics, average number of paid claims per child, and average cost per child for each child with and without OFC continuously enrolled in Medicaid each year of life through age four. For children with OFC, these analyses also included cleft type and presence of other anomalies. Chapter Three examines the timeliness of services in accordance with the ACPA guidelines among children with OFC who were continuously enrolled in the first and

second year of life. The research questions, hypotheses, and analyses employed for all specific aims are summarized in Tables 1.4-1.6.

Table 1.4. Aim 1: To describe health care service use and costs among children with and without orofacial clefts on Medicaid during the first five years of life

Research Question	Hypothesis	Analysis
What are the differences in specific health care service use and cost by service type such as medical, inpatient/hospitalization, outpatient, mental health, home health, dental, and well-child care between cases and controls for each year of life?	Cases will differ significantly from controls with regards to health service utilization and costs. Medical, inpatient, outpatient and dental services will be the most utilized services and most costly services among cases due to their complex medical needs. Medical, inpatient, and outpatient services will be most utilized during the first year of life among the cases and controls, thereby having higher costs for these services in the first year of life.	Descriptive statistics
What are the differences in cumulative health service use and costs between cases and controls for each year of life?	Cases will differ significantly from controls with regards to total health service utilization and costs, especially during the first two years of life.	Descriptive statistics
Among cases, what are the differences in specific health care service use and costs by cleft type and presence of other anomalies for each year of life?	Children with non-isolated OFC will have the highest frequency of medical services and will have the highest costs. Medical, inpatient, and outpatient services during the first two years of life will be utilized the most while other health services will be used during the later years of a child's life. Over the five-year period, children with non-isolated OFC will have the highest medical and dental costs.	Descriptive statistics

Table 1.5. Aim 2: To determine the effects of individual and system characteristics on health service utilization and costs among children with orofacial clefts and unaffected children on Medicaid during the first year of life

Research Question	Hypothesis	Analysis
What are the effects of maternal, child, and system characteristics on health care service utilization among cases and controls?	Among cases and controls, minority mothers who are younger, less educated, not married, have more children, not participating in WIC, and received no PNC will have less total health care service utilization due to being less familiar with the health care system and increased barriers to accessing health care. Children who are younger with non-isolated OFC and participating in WIC will have greater total health care service use.	Bivariate and multivariate log-linear regression for each service category
What are the effects of maternal, child, and system characteristics on health care costs among cases and controls?	Children without OFC whose mothers are of racial/ethnic minority, younger, less educated, not married, have more living children, on WIC, and did not receive PNC will have lower Medicaid costs. Children who are older with non-isolated OFC and not participating in WIC will have higher Medicaid costs.	Bivariate and multivariate, specifically 2-part model: 1) binary logit; and 2) ordinary least squares regression for each cost category

Table 1.6. Aim 3: To assess the timeliness of certain services according to the American Cleft Palate-Craniofacial Association recommendations among children with orofacial clefts

Research Question	Hypothesis	Analysis
What proportion of children with CP or CLP received primary surgery within the first 18 months of life?	A large majority of children with isolated CP or CLP will have received their primary cleft surgery by 18 months of life.	Descriptive statistics
What proportion of children with CL received primary cleft surgery within the first six months of life?	A large majority of children with isolated CL will have received their primary cleft surgery within six months of life.	Descriptive statistics
What maternal, child, and system characteristics are associated with timely receipt of primary cleft surgery?	Children with isolated CL or isolated CP will be more likely to receive timely cleft surgery than children with non-isolated CLP. Families of children with OFC traveling $\leq$ 30 minutes to a craniofacial center or team will be more likely to receive timely primary cleft surgery than families traveling $\geq$ 90 minutes.	Geocoding, geographic information systems, and multivariate logistic regression
What proportion of children with OFC received otolaryngologic care within the first six months of life?	Over 50% of children with OFC will have received otolaryngologic care within the first six months of life.	Descriptive statistics

Research Question	Hypothesis	Analysis
What proportion of children with OFC received audiological and dental services and speech and language therapy within the first year of life?	About 30% of children with OFC will have received audiological and dental services and speech and language therapy within the first year of life.	Descriptive statistics
What proportion of children with OFC received genetic services within the first two years of life?	Less than 10% of children with OFC will have received genetic services within the first two years of life.	Descriptive statistics

Human Subjects Concerns

This dissertation research involved secondary data analysis. The primary human subjects concern for this research was maintaining confidentiality of the data. The data were stored separately in a locked filing cabinet and were password protected. All the data sets was stored on a secure network server at the State Center for Health Statistics. All staff at the State Center for Health Statistics, including all NCBDMP staff, are required to sign certificates of confidentiality. This dissertation research was approved by the University of North Carolina at Chapel Hill Public Health Institutional Review Board, North Carolina Division of Public Health Institutional Review Board, and the North Carolina Division of Medical Assistance.

CHAPTER II

PATTERNS AND PREDICTORS OF HEALTH CARE SERVICE UTILIZATION AND MEDICAID COSTS AMONG CHILDREN WITH AND WITHOUT OROFACIAL CLEFTS DURING THE FIRST YEAR OF LIFE IN NORTH CAROLINA, 1995-2002

Abstract

Background and Objectives: Orofacial clefts (OFC) are the third most common birth defect in the United States. National data that examine health service use and costs of children with special health care needs are limited, and only a few studies address children with OFC. This study examines patterns and predictors of health service use and costs among children with and without OFC during the first year of life.

Methods: Data from the North Carolina BabyLove files, birth defects registry, and Medicaid enrollment and paid claims were linked to identify resident children born 1995-2002 with OFC (cases, N=565) and without OFC (controls, N=5,674) who were continuously enrolled in Medicaid during infancy. The average number of paid claims and average cost per child for cases and controls were determined for medical, inpatient, outpatient, dental, well-child care, mental health, home health, and total. The rate of service utilization and cost in each category was compared among cases and controls by calculating utilization and cost ratios. Bivariate analysis was also conducted to determine any differences in maternal, child, and system characteristics among cases and controls. Poisson multivariate regression analyses were employed to assess the effect of selected maternal, child, and system characteristics on health service use among cases and controls. To examine the effect of these same

characteristics on health care costs among cases and controls, two-part models were conducted. Likelihood ratio and Chow tests were conducted on each model to determine if the associations were similar for cases and controls.

Results: During the first year of life, mental and home health services were utilized almost 20 times more by cases than controls, and the mean cost per child with OFC for mental and home health services was 37 and 45 times higher respectively compared to a child without OFC ($632 vs. $17 and $1,843 vs. $41, respectively). In infancy, the average total health care cost per child with OFC was six times the average total cost per child without this condition ($22,642 vs. $3,900). The total cost for children with OFC on Medicaid was $12,792,634 compared to $2,212,839 for children without OFC, which were randomly sampled in a 1:1 ratio. Characteristics associated with greater health service use cost varied across service use and cost categories and among cases and controls.

Conclusions: This study confirms that children with OFC utilize significantly more services and have significantly more Medicaid costs than children without OFC. These results provide accurate and current data on service utilization and costs of children with OFC for service and program planning and policy development. Targeting care coordination and early intervention should be investigated as a means for decreasing long-term costs associated with treating these children.

Background

Several studies have examined health service utilization and costs among children with and without special health care needs (20, 21, 26, 30, 65-95, 155). Previous studies have documented that CSHCN utilize services more and thus have higher medical costs than

children without special health care needs. These studies also have shown that service use and expenditures for CSHCN vary considerably across different chronic condition categories and across different expenditure categories such as inpatient, physician, and outpatient services (20, 21, 70). In addition, these studies have shown disparities in access and utilization among children with and without special health care needs (26, 88, 95-98), and CSHCN from less educated families and non-Hispanic Black CSHCN were significantly less likely to use many medical and health-related services (26). However, these previous studies and national data on CSHCN from the Healthcare Cost and Utilization Project, Medical Expenditure Panel Survey, and the National Survey of CSHCN are limited because they do not adequately control for factors like child's age, condition such as birth defect type, and presence of other birth defects that could have influenced utilization and thereby costs (16, 18-30, 77, 93, 95, 103). These factors are critical because patterns of medical and health-related service use and costs for CSHCN such as children with birth defects can differ considerably by biological, familial, social, and developmental factors (20, 31).

Some studies have examined health service use and expenditures among children with birth defects, including OFC (16-20, 70, 101-103, 156). The two most comprehensive studies conducted on costs of birth defects was conducted by Waitzman et al. in 1994 and Harris and James in 1997 (18, 19). Waitzman et al. found that the per capita medical costs of infants with OFC born in 1988 in 1992 dollars was $6,794 (16-19). Two recent studies used 2003 and 2004 data from the Healthcare Cost and Utilization Project to examine hospitalizations and costs in children with birth defects in the United States. These authors found the mean hospital charge for children with OFC was $15,397 per hospitalization and total hospital charges were almost $54 million during this time period (157). While providing

important health service use and cost data on children with birth defects, these two studies only examined hospitalizations using hospital discharge data. One of these studies only examined costs during the newborn period (102, 103). All the previous studies on health service use and cost of children with birth defects did not adjust for key variables such as insurance, place of residence, or the child's condition and only examined service use and cost for children with isolated CP and children with CL with or without CP (102, 103, 156, 158).

A few studies have focused solely on costs of children with craniofacial conditions such as OFC and costs of particular treatments like nasoalveolar molding. From a 1994 birth cohort, Berk et al. found over the first five years of life, children with OFC on average were billed $4,046 for children with CL, $3,610 for children with cleft CP, and $10,330 for children with CLP for services rendered at the University of Pittsburgh Cleft Palate-Craniofacial Center (31). However, this was a small study only one clinic. The costs examined in this study were fees billed to patients or charges and not the actual amount paid for the services. Charges have been shown to be a proxy for cost, but bear little resemblance to actual costs (104). Other studies examining costs associated with OFC have focused on only certain aspects of treatment like nasoalveolar molding, gingivoperiosteoplasty, and alveolar bone graft, and/or specifically addressed one type of cleft type such as unilateral cleft alveolus or complete unilateral cleft lip and palate (25, 106, 107, 159). These studies were also small and did not control for any important variables such as maternal age, education and race that could also be related to health care costs among children with OFC.

Comprehensive and contemporary data on health care service utilization and costs are lacking for children with birth defects, especially for children with OFC in North Carolina. To date, no study has examined patterns and predictors of health service use and costs in

children with birth defects such as OFC. Moreover, no data exists on health service use and costs of children with isolated CL, children with isolated OFC, and children with OFC and other birth defects. Identifying trends and characteristics associated with health service use and costs among children with and without OFC and among children with OFC by cleft type and presence of other birth defects will provide much needed information on this population. The objective of this study was to compare the patterns of health care service use and costs among children with OFC and unaffected children, and the variability among children with OFC by cleft type and presence of other anomalies. A secondary objective was to examine the effect of selected maternal, child, and system characteristics on certain categories of health service use and cost among both groups of children.

Methods

Study Design and Sample

This retrospective, case-control study included children born in North Carolina between January 1, 1995, and December 31, 2002, who were continuously enrolled in Medicaid during infancy. Cases were live births diagnosed with an OFC and ascertained by the NCBDMP using British Pediatric Association codes 749.000-749.290. Controls were selected in 10:1 ratio from a simple random sample of Medicaid eligible children born during the same time period from the BabyLove files. This ratio was chosen to ensure greater than 95% power ($\alpha = 0.05$) to detect differences in service use and costs and sufficient numbers for any subgroup analysis. Because the probability of selecting children with OFC or other major birth defects was less than 3.0%, these children were not excluded upfront from the

random selection of controls. Children who died within the first 12 months of life, who were born out of state, or who were adopted were excluded from the study.

Using the infant Medicaid identification number, both cases and controls were matched from the BabyLove files to the Medicaid enrollment records to identify children who were continuously enrolled in Medicaid during the first year of life. The matching rate of the BabyLove records to the Medicaid enrollment records was 98.5%. Continuous enrollment in Medicaid was defined as enrollment greater than or equal to 11 months in the first year of life. This is a commonly used definition for continuous enrollment in Medicaid and utilized as the standard definition for the Health Plan Employer Data and Information Set, which is the most widely used set of performance measures in the managed care industry (66, 144, 145). Enrollment records were then matched to the Medicaid paid claims to determine health care service utilization and costs. The matching rate was 100%.

Data Sources Utilized and Variable Construction

Data sources for this study included the North Carolina BabyLove files, NCBDMP, and Medicaid enrollment records and paid claims. The BabyLove files are composite North Carolina birth files matched with Medicaid newborn hospitalization records, maternal delivery records, and maternity case management records and contain information on child service coordination, WIC, Medicaid status, PNC source, MCC services, Medicaid newborn costs in the first 60 days of life, and Medicaid infant costs in the first year of life. The NCBDMP is a population-based, statewide program that utilizes passive and active ascertainment methods and includes all resident live births, fetal deaths, and therapeutic abortions at any gestational age. The program covers all of North Carolina hospitals except

military hospitals and approximately 120,000 resident births per year. Each health care service and cost category was tracked using Medicaid paid claims. Records of all services received and paid for by Medicaid for calendar years 1995 through 2004 were extracted to allow for at least one year of health service utilization and costs for all children born during the study period.

The primary outcomes of interest were several health service utilization and cost categories, which included medical, inpatient, outpatient, mental health, home health, dental, well-child care, and total. These categories have been employed in previous studies on health service use and costs (21, 28, 66, 77, 95, 145). Except for mental health, all data elements for health service use and costs were obtained directly from categories on the Medicaid paid claims. The mental health category was created using service categories, provider specialty codes (also known as billing provider specialty codes), provider type codes, diagnostic related group codes, and procedural codes that referred to mental retardation, developmental disabilities, and substance abuse services. The construction of the mental health variable was in consultation with the North Carolina Division of Mental Health**,** Developmental Disabilities and Substance Abuse Services**.** Medical paid claims were defined as any professional claims or fees associated with office based physicians' appointment(s) as well as professional claims associated with any outpatient and inpatient visit. Inpatient paid claims included any hospital and facility fees such as room use, labs, and X-rays associated with an inpatient visit. Outpatient paid claims consisted of any hospital and facility fees associated with an outpatient visit. Outpatient claims also included any emergency department claims. "Other" claims included professional crossover, medical vendor, and outpatient and inpatient crossover paid claims. Total paid claims consisted of the

sum of paid claims from all the service categories, and total costs were defined as the sum of costs from all the cost categories. In this study, the number of paid claims in each health service category was a proxy for health service utilization.

Several factors that might affect health service use and cost were also investigated. These variables were categorized into maternal, child, or system characteristics. Maternal characteristics included age, number of living children, race/ethnicity, education, marital status, and initiation of PNC in the first trimester. Child characteristics included birth weight, preterm birth, and gender. For children with OFC, child characteristics also included cleft type and presence of other anomalies. Cleft type was stratified as CL alone, CP alone, or CLP. Presence of other anomalies included isolated or multiple anomalies. An isolated anomaly was defined as an OFC diagnosis only, and multiple anomalies were defined as a diagnosis of OFC and another birth defect. System characteristics consisted of source of PNC, receipt of MCC services, receipt of WIC, birth hospital level of care, perinatal care region, and place of residence. Perinatal care regions were administratively and geographically defined as northwestern, northeastern, southwestern, southeastern, western, and eastern. These regions were established in the 1980's in an effort to develop regional referral networks of perinatal care in North Carolina. For place of residence, Urban Influence Codes were utilized. These codes are based on counties and classified into 12 levels that build on the Office of Management and Budget metropolitan and nonmetropolitan dichotomy (150-154). Adjacency includes physical adjacency and at least 2% of the population commuting to the metropolitan or micropolitan area (154). These codes are based on two main categories: metropolitan and nonmetropolitan counties. In this study, the 12

levels were collapsed into four categories based on metropolitan, nonmetropolitan, adjacency, and non-adjacency areas.

Statistical Analysis

Relationships between maternal, child, and system characteristics and cases and controls were assessed using chi-square tests. To examine differences in service use and cost categories between children with OFC and unaffected children, service utilization and cost ratios for medical, inpatient, outpatient, home health, mental health, dental, well-child care, other, and total were constructed. These ratios were defined as the average number of paid claims or mean cost per child with OFC divided by the average number of paid claims or mean cost per child without OFC respectively. These ratios were evaluated for statistical significance using the Wilcoxon rank sum test, which is commonly applied to data that exhibit skewness. A p-value less than 0.05 for both the Wilcoxon rank sum test and chi-square test were considered statistically significant.

In the multivariate analysis, all maternal, child, and system characteristics were included in the models on the basis of contributing significantly to the model as indicated by likelihood ratios tests where the cut point was p less than 0.05. Variables were also included on the basis of *a prior* hypothesis according to Andersen's Behavioral Model of Health Service Utilization and/or as indicated in previous literature as discussed in Chapter One. For each child, the total count of paid claims and total costs for a particular service such as mental health during the first year of life were tabulated. All covariates were included in the model, and for all the categorical variables, indicator variables were created. Multivariate regression models were run separately on children with and without OFC. Due to insufficient

data for well-child care, dental, and other health service and cost categories for cases and controls, these categories were not examined in the multivariate analyses.

To examine how selected maternal, child, and system characteristics were associated with the number of paid claims (i.e., service use) for each health service category, log-linear Poisson regression was employed. Poisson regression is a form of the generalized linear model and, in this case, models the log of count ratios or number of events. Beta coefficients, standard errors, effect estimates and 95 percent CI were computed for each covariate to assess the magnitude and precision of the effect estimates. However, only the count ratios, which are the effect estimates, and 95 percent CI are presented. The specific outcomes modeled were medical, inpatient, outpatient, mental health, home health and total.

To determine how selected maternal, child, and system characteristics were associated with each cost category, a two-part model was employed for each outcome. A two-part model was appropriate for the cost categories because a disproportionate number of children had zero costs, which were the nonusers of service. First, health care costs for each child for each outcome were created into a binary variable. Then, a binary logit model was used to examine the effect of selected characteristics on each cost category to determine the likelihood of incurring any costs by the selected covariates. Odds ratios were computed to determine the magnitude of these relationships, and 95 percent CI were generated to examine the precision of these effect estimates. If a cost category had less than five percent of children who did not have costs, then a logit regression was not conducted only ordinary least squares regression analysis, which is the second part of the two-part modeling. Because outpatient, mental health, and home health cost categories had greater than five percent of

children with zero costs, these categories were the only ones examined in the binary logit analyses.

Likelihood ratio tests were employed for each cost category to determine if each regression coefficient for maternal, child, and system characteristics in each cost category were similar for cases and controls. Likelihood ratio tests were used due to the non-normally distributed errors. Non-normally distributed errors can be a result of heteroscedasticity and skewed data, which is commonly exhibited with expenditure data.

For the second part of the two-part model, ordinary least squares regression analyses were employed conditional on those children who had costs. Using a continuous variable for each outcome, all costs were log transformed to reduce skewness. Beta coefficients, standard errors, effect estimates, and 95 percent CI were obtained to determine the magnitude and precision of the effect estimates, which were cost ratios. However, for ease of interpretation, only cost ratios and 95 percent CI are reported. The specific outcomes modeled were medical, inpatient, outpatient, mental health, home health and total costs.

Chow tests were employed to determine any differences between the models for cases and controls in each cost category. The Chow test is an analysis-of-variance F-ratio test, which tests for structural change in regression analysis. Specifically, it jointly tests the intercept and slope of the dummy variables. Because the Chow test assumes homoscedasticity, it was not used to compare the regression coefficients for cases and controls in the binary logit models.

All analyses were conducted on children with and without OFC who were continuously enrolled during the first year of life using SAS software, version 9.1. This study was approved by the University of North Carolina at Chapel Hill Public Health Institutional

Review Board, North Carolina Division of Public Health Institutional Review Board, and the North Carolina Division of Medical Assistance.

Results

Between 1995 and 2002 in North Carolina, 1,355 children were born with OFC. Of these children, 103 died during the first year of life and were excluded from the analysis. Of the remaining 1,252 children with OFC, 50.1% (N=627) were on Medicaid. The original control sample size was 6,270 children on Medicaid. After excluding controls that had a diagnosis of OFC, other major birth defect(s), or died during the first year of life, the final control sample size was 6,127 children. From 6,754 children with and without OFC, 69 children had missing data for one or more of the variables analyzed. Because this represented less than 2.0% of the entire dataset and had very little effect on the results, they were not deleted from the data set; however, they were deleted in the analyses. Ninety percent (N=565) of children with OFC and 92.6% (N=5,674) of children without OFC were continuously enrolled in Medicaid during the first year of life, which yielded the final sample size of 6,239 children.

Sample Population Characteristics

Table 2.1 indicates the distributions of maternal age and education and number of living children were similar among both groups of children. However, 15.2% more mothers of children with OFC were White/non-Hispanic compared to mothers of children without OFC. Conversely, 14.3% more mothers of children without OFC were Black/non-Hispanic compared to mothers of children with OFC. Children with OFC were significantly more likely to be born LBW or preterm than children without this condition. Children with OFC

were less likely to be female than children without this condition. Children with multiple anomalies was more common among children with isolated CP (52.3%) than children with isolated CL (10.7%) and children with CLP (37.1%). Among children with OFC, more children had isolated CL and CLP than isolated CP. Mothers of children with and without OFC were similar in the receipt of PNC and MCC services. The percentages of mothers receiving MCC services, WIC, and PNC at a health department were higher among study participants than all mothers giving birth in North Carolina during this time period.

Health Service Use

Table 2.2 shows the average number of paid claims per child for cases and controls, corresponding ranges, and utilization ratios. Overall, children with OFC utilized health services 1.7 times more than children without OFC during the first year of life. The utilization ratios for each service category except well-child care were significantly higher. The mean number of paid claims per child with OFC for mental and home health services was almost 20 times higher compared to a child without OFC. Compared to controls, the mean number of paid claims per child with OFC for medical and inpatient service use was about two times higher and for outpatient services it was 2.7 times higher. For dental and well-child care services, the mean number of paid claims per child was low for children with and without OFC (0.2 vs. 0.0 and 3.7 vs. 3.8, respectively).

Among children with OFC, Table 2.3 reveals that the average number of paid claims per child with CP and CLP were similar in all service categories compared to the average number of paid claims per child with CL. Children with OFC and another birth defect had about twice the average number of medical, inpatient, outpatient, mental health, and total

paid claims per child compared to a child with OFC only. Compared to children with isolated anomalies, the mean number of paid claims for home health services per child with multiple anomalies was 11.4 times higher (Table 2.4).

Figures 2.1 and 2.2 represent the percentages of each service category of total service use for cases and controls. Medical, inpatient, and outpatient paid claims contributed similarly to the composition of total service use for cases and controls, but the composition of dental, mental health, and home health differed. Mental health and home health paid claims together comprised less than 5.0% of the total paid claims for children with OFC whereas these paid claims comprised less than 0.5% for children without this condition. In both groups of children, mental health and home health services were utilized less than medical, inpatient, and outpatient services. For children without OFC, well-child care paid claims comprised 3.8% more of total paid claims than children with OFC.

Predictors of Health Service Use

After adjusting for all the covariates, the effect of selected maternal, child, and system characteristics on the number of paid claims (i.e., health care service utilization) varied among cases and controls and each health service category. For children with OFC, no one characteristic was associated with higher service utilization in every service category. In contrast, unaffected children who were male and born LBW had significantly greater service use in all service categories compared to female children without OFC who were not born low birth weight. Children with and without OFC who were born LBW utilized significantly more medical, inpatient, and home health services, and had greater total service use than children were not born LBW. Unlike children with OFC, children without OFC who were

born preterm had significantly greater service use in all service categories except mental health. Children with OFC and who were born preterm had 167% greater mental health service use, but had 54% less home health service use than children with OFC who were not born preterm. Among children with and without OFC, children residing in the northeastern and western region of the state had significantly greater medical and total service use compared to children living in the northwestern part of the state. In both groups of children, children living in noncore adjacent and noncore areas not adjacent to a metropolitan or small town had greater mental health service use than children living in metropolitan areas. Also, for both groups of children, mothers who received MCC services had significantly greater medical and total service use than mothers who did not receive such services (Appendix A Tables 1-6 and Appendix B Tables 1-2).

When cleft type and presence of other anomalies variables were added to the multivariate regression models for health service use, the effect estimates for the other characteristics remained stable. These models indicated that children with CP or CLP and children with multiple anomalies were positively associated with health service use in all service categories. Children with CP had 22% greater total service use and children with CLP had 38% greater total service use than children with CL. Children with OFC and another birth defect had 64% greater total service use than children with OFC only (Appendix C Tables 1-6). Because health care costs can be a proxy for health service use and have important policy implications, the rest of this chapter focuses on the effect of selected individual and system characteristics on health care costs among children with OFC and unaffected children

Health Care Costs

The average cost per child with OFC was six times the total cost compared to the average cost per child without OFC during the first year of life, $22,642 vs. $3,900 respectively (Table 2.5). During infancy, the total cost for children with OFC on Medicaid was $12,792,634 compared to $2,212,839 for a 1:1 random sample of children without OFC. The mean mental health and home health cost per child with OFC was 37 and 45 times higher respectively compared to a child without OFC. Relative to controls, the mean outpatient cost per child with OFC was 16 times higher. The mean dental cost per child was low for children with and without OFC, $13 and $0 respectively (Table 2.5). The costs ratios for each cost category except well-child care were statistically significant.

Children with isolated CL and children with CLP had similar average costs per child in each service category during the first year of life. Compared to the total average cost of a child with isolated CL, the total average cost of a child with isolated CP or CLP was significantly higher (Table 2.6). During the first year of life, the total cost of children with isolated CP and children with CLP were similar, $5,307,782 and $6,623,764 respectively. The total cost of children with isolated CL was significantly less, $861,088.

The average total cost of a child with OFC and another birth defect was almost five times that of a child with OFC only. The average cost per child with multiple anomalies for home health services was 36 times higher than for a child with an isolated anomaly (Table 2.7). The total cost for children with multiple anomalies was $9,076,183 compared to $3,716,451 for children with OFC only.

Figures 2.3 and 2.4 represent the percentage of costs by category of total costs for children with and without OFC. Inpatient costs contributed similarly to the composition of total costs

for cases and controls, but other service categories contributed differently to total costs. Outpatient costs contributed almost 10% more to the composition of total costs for children with OFC than for children without OFC. Conversely, medical costs contributed almost 9% more to the composition of total costs for children without OFC than for children with OFC.

Predictors of Health Care Costs

Binary Logit Results for the Effect of Selected Individual and System Characteristics on Any Outpatient, Mental Health, and Home Health Costs

Children with OFC whose mothers who were 25 years or older, had less than a high school education, received MCC services, and resided in the southeastern region of the state were significantly more likely to have outpatient costs than their counterparts. In comparison, these maternal and system characteristics were not positively associated with any outpatient costs among children without OFC. Children with OFC who resided in the southwestern, southeastern, and western regions of the state were two to three times as likely to have any home health costs compared to children with OFC living in the northwestern perinatal care region. Perinatal care region was not positively associated with any home health costs among children without OFC (Tables 2.8-2.10 and Appendix B Tables 3 and 4).

When cleft type and presence of other anomalies variables were added to the multivariate models for children with OFC, the effect estimates for the other characteristics remained stable. Children with CP or CLP and children with multiple anomalies were positively associated with any mental health and home health costs. Children with CLP and children with multiple anomalies were significantly more likely to have outpatient costs than children with CL only and children with OFC only (Appendix C Table 7).

For both children with and without OFC who were born LBW or preterm and whose mothers received PNC in the second or third trimester or MCC, and resided in noncore areas adjacent and nonadjacent to a metropolitan area or small town were significantly more likely to have mental health costs than their counterparts. For children with and without OFC born LBW or born at a tertiary care center were more likely to have home health costs than children not born LBW or born in a community hospital. Children without OFC born preterm were 3.4 times more likely to have home health costs than children without OFC who were not born preterm (adjusted OR: 3.43; 95% CI: 2.10, 5.59) (Table 2.10). Likelihood ratio tests indicated that the regression coefficients for the likelihood of outpatient, mental health, and home health costs for children with OFC were statistically different from unaffected children (p less than 0.0001). As indicated in Tables 2.8-2.10, in general, the effects of selected individual and system characteristics on any outpatient, mental health, and home health costs varied among the cost categories and among children with and without OFC.

Ordinary Least Squares Regression Results for the Effects of Selected Individual and System Characteristics on Medical, Inpatient, Outpatient, Mental Health, Home Health, and Total Costs among Children who had Costs

Medical Costs. After adjusting for maternal, child, and system characteristics, Table 2.11 indicates that among children with medical costs, children with OFC whose mothers were greater than 30 years old or who were Black/non-Hispanic had significantly less medical costs than mothers 21-24 years old or White/non-Hispanic mothers. Children with OFC who were born at a tertiary care center had 26% greater medical costs than children with OFC born in a community hospital.

In children without OFC who had costs, mothers who had less than a high school education had significantly greater medical costs than mothers who were 21-24 years old and

mothers who were Hispanic had significantly less medical costs than mothers who were White/non-Hispanic.

In both groups of children who had costs, children who were born LBW or preterm had greater medical costs than children who were not born LBW or preterm. Children with OFC who were born LBW had 34% greater medical costs and children without OFC who were born LBW had 61% greater costs than children who were not born LBW. In children with and without OFC, receiving MCC was associated with significantly higher infant medical costs (Table 2.11 and Appendix B Table 6 and 7).

Inpatient Costs. Table 2.12 indicates that among children with OFC who had inpatient costs, mothers who were greater than 30 years old had 30% less inpatient costs compared to mothers who were 21-24 years old. Mothers of children with OFC who received MCC had significantly higher inpatient costs than mothers who did not receive MCC. Children with OFC who resided in the southeastern, eastern, western, or noncore areas not adjacent to a metropolitan area or small town had greater inpatient costs than children residing in the northwestern perinatal care region or a metropolitan area of the state. In comparison, among children without OFC, children residing in the northeastern and eastern regions of the state had significantly higher inpatient costs than children residing in the northwestern area of the state.

Like with medical costs, in both groups of children, children who were born LBW or preterm had significantly higher inpatient costs than children who were not born LBW or preterm. Children with OFC born LBW had 96% greater inpatient costs, and children without OFC born LBW had 152% greater inpatient costs than children not born LBW. Similarly, children with OFC born preterm had 77% greater inpatient costs, and children without OFC

born preterm had 101% greater inpatient costs than children not born preterm. Children with OFC and unaffected children born in tertiary care centers had significantly higher inpatient costs than children born in community hospitals (Table 2.12 and Appendix B Tables 6 and 7).

Outpatient Costs. Among children with OFC that had outpatient costs, mothers who were less than 20 years old and 25 and older had substantially less outpatient costs than mothers who were 21-24 years old. Among children with OFC, children residing in the southwestern, western, and any other area besides a metropolitan area were associated with less outpatient costs compared to children living in the northwestern region and metropolitan areas. Children with OFC living in the western area of the state had 76% less outpatient costs than children living in the northwestern region. For children without OFC, being born LBW, born in a tertiary care center, and whose mothers received PNC somewhere other than at a local health department and received WIC were associated with significantly higher outpatient costs than their counterparts. Among children with OFC, all of these factors except LBW were also associated with lower outpatient costs than their counterparts (Table 2.13 and Appendix B Tables 6 and 7).

Among both children with OFC and unaffected children, mothers who had less than a high school education had higher outpatient costs than mothers who had greater than a high school education. However, children with OFC whose mothers had less than a high school education had 56% greater outpatient costs, and children without OFC had 15% greater outpatient costs. Children with and without OFC living in the northeastern region had higher outpatient costs than children living in the northwestern region, 22% and 37% respectively. Children with OFC living in the eastern region had 52% lower outpatient costs and unaffected children

had 16% lower outpatient costs than children living in the northwestern region (Table 2.13 and Appendix B Tables 6 and 7).

Mental Health Costs. Table 2.14 indicates that children with OFC whose mothers were Black/non-Hispanic had 40% greater mental health costs than mothers who were White/non-Hispanic. In comparison, children without OFC whose mothers were Black/non-Hispanic had 33% less mental health costs than mothers who were White/non-Hispanic. Children with OFC who were born LBW had 50% less mental health costs than children with OFC not born LBW. Conversely, children without OFC who were born LBW had 17% greater mental health costs compared to children not born LBW. Children with OFC born preterm had 88% greater mental health costs than children not born preterm. Being born preterm was not associated with greater mental health costs among children without OFC. Children with OFC whose mothers did not receive PNC at a local health department had 43% less mental health costs compared to children whose mothers received PNC at a local health department. Children without OFC whose mothers did not receive PNC at a local health department had 87% greater mental health costs compared to mothers who received PNC at a local health department. Children with OFC residing in the southwestern region of the state had 54% less mental health costs whereas children without OFC had 29% greater mental health costs than children residing in the northwestern region of the state. Among children with and without OFC, Hispanic mothers, mothers who received WIC, and mothers who lived in the northeastern region of the state had less mental health costs than their counterparts (Table 2.14 and Appendix B Tables 6 and 7).

Home Health Costs. Among children with OFC who had home health costs, children whose mothers were less than or equal to 20 years old had 159% greater home health costs

and mothers who were 25-29 years old had 61% greater home health costs than mothers who were 21-24 years old (Table 2.15). In comparison, maternal age was not associated with higher home health costs among children without OFC. Living in a noncore adjacent or not adjacent to a metropolitan or small town was negatively associated with home health costs among children with OFC whereas this was positively associated with home health care costs among unaffected children. Among children without OFC, mothers who had less than or equal to a high school education had lower home health costs; however, mothers with less than a high school education was not associated with decreased home health costs among children with OFC (Table 2.15 and Appendix B Tables 6 and 7).

Among both groups of children, Hispanic mothers and mothers who did not receive PNC at a local health department had less home health costs than White/non-Hispanic mothers and mothers who did receive PNC in a local health department. Children born LBW, mothers who received maternal care coordination, and who resided in the northeastern and western part of the state were associated with greater home health care costs than their counterparts (Table 2.15 and Appendix B Tables 6 and 7).

Total Costs. Among children with OFC, mothers who were Black/non-Hispanic or Hispanic had about 20% less total costs but mothers of "other" minority race had 25% greater total costs than mothers who were White/non-Hispanic. Maternal race was not associated with increased or decreased total health costs among children without OFC. Among children with OFC, mothers who received MCC had 24% greater total costs than mothers who did not receive MCC, but this was not statistically significant. In comparison, among children without OFC, mothers who received MCC had 70% greater total costs, which was statistically significant. Unlike children without OFC, children with OFC who

resided in noncore adjacent areas had significantly less total costs than children living in metropolitan areas. Among both groups of children, mothers who had less than a high school education and children who were born LBW or preterm had higher total costs than their counterparts (Table 2.16 and Appendix B Tables 6 and 7).

Chow tests indicated that all the ordinary least squares regression models for children with OFC were statistically different from the models for children without OFC (p less than 0.0001). Nevertheless, some maternal, child, and system characteristics were consistently associated with lower or higher costs in most of the cost categories among children with and without OFC who had costs. Among children with OFC, mothers who were greater than or equal to 30 years old had lower medical, inpatient, outpatient, and mental health costs compared to mothers who were 21-24 years old. Among children without OFC, mothers who had less than a high school education had significantly higher medical, inpatient, outpatient, and total costs compared to mothers with greater than a high school education. Residing in the northeastern region of the state was associated with significantly higher inpatient, outpatient, home health and total costs among children without OFC. In both groups of children, children who were born LBW or preterm were associated with greater medical, inpatient, home health and total costs. In three of the cost categories, mothers who received MCC and children who were born at a tertiary care center were associated with higher costs among children with and without OFC. However, these categories were not necessarily the same for children with OFC and unaffected children (Tables 2.11-2.16 and Appendix B Tables 6 and 7).

When cleft type and presence of other anomalies were added to the multivariate regression model for children with OFC who had costs, the effect estimates for the other maternal, child,

and system characteristics remained stable. Children with CP had significantly greater inpatient costs but had significantly less outpatient costs than children with CL. Children with CLP had significantly greater medical, inpatient, outpatient, and total costs than children with CL. For example, children with CLP had 33% more medical costs than children with CL. Children with multiple anomalies had significantly higher costs in all cost categories (Appendix C Tables 8-13). For instance, children with multiple anomalies had 122% greater total costs than children with OFC only (Appendix C Table 13).

Discussion

This study found a child with OFC had an average of $18,742 greater Medicaid costs than a child without OFC during the first year of life. Children with OFC cost more than $10 million to Medicaid than children without this condition in the first year of life. This study also confirms that children with OFC utilize significantly more health services and have significantly more health care costs than children without OFC, especially for medical, inpatient, outpatient, mental health, and home health during infancy. While this was expected, the magnitude of the difference was previously unknown. Mental and home health services had the highest utilization ratios, about 20 times more for children with OFC than children without OFC. Mental and home health costs also had the highest cost ratios, about 37 and 45 times more respectively for children with OFC than children without this condition. The average number of dental paid claims and average dental cost per child for both cases and controls were also extremely low, 0.2 and 0.0 and $13 and $0 respectively.

This study also found that the effect of maternal, child, and system characteristics on health service utilization and cost tended to vary among each category and among children with and without OFC. However, among children who had health care costs, children who

were born LBW was associated with higher medical, inpatient, home health, and total costs among cases and controls. Children with and without OFC who were born preterm were also associated with higher medical, inpatient, and total costs.

For children with OFC, it was hypothesized that these children would have significantly higher service use and cost than children without this condition, especially with medical, inpatient, and home health services. It was also hypothesized that children with CP or CLP and children with multiple anomalies would significantly utilize services more and would have significantly higher costs than children with CL and children with isolated clefts. These results substantiate these hypotheses. It was also assumed that younger, less educated mothers and Black/non-Hispanic and Hispanic mothers would be inversely associated with all service use and cost categories. For some service and cost categories, there was a slight or no association between younger, less educated mothers and medical, inpatient, and total service use and costs among children with and without OFC. Yet, there was a strong positive association such as with less educated mothers and greater outpatient service use and cost among both groups of children. This may have been due to the use of emergency room care as a primary source of care because previous studies have indicated lower socioeconomic families often have no regular source of care (83, 98).

In this study, children with OFC were significantly more likely to be born LBW or preterm than children without this condition. This was to be expected as children with OFC are at an increased risk of being born LBW and/or preterm, especially if they have another birth defect as well (160, 161). This study also found that having multiple anomalies was more common among children with CP, which was to be expected (161). The gender proportions of children without OFC were representative of all live births during this time

period compared to children with OFC. This was to be expected as male children with OFC are affected more frequently by non-isolated CL with or without CP than female children.

We also found that the percentages of less educated mothers and mothers receiving MCC services, WIC, and PNC at a health department were higher among study participants than all mothers giving birth in North Carolina during this time period. This was to be expected among mothers receiving Medicaid for several reasons such as mothers can only receive MCC if they are Medicaid-eligible, and this population was of relatively low-income.

With regards to maternal race, the percentage of White/non-Hispanic mothers in the control group was about 13 percentage points less and the percentage of Black/non-Hispanic mothers of children with OFC was about 15 percentage points less than the general population of White/non-Hispanic and Black/non-Hispanic mothers on Medicaid during the study period. The percentage of "Other" race/ethnic category in both groups of children was comparable to the general population of mothers on Medicaid in the state during the study period.

Results from this study may be due to several factors such as children with OFC are at an increased risk for being born LBW and parental perception of need. Children with OFC are at an increased risk for being born LBW, which thereby increases the risk of developmental disabilities. Previous studies are inconsistent as to whether children with OFC have an increased risk of being born preterm as well. Nevertheless, children with OFC born both preterm and LBW often have prolonged hospitalizations and secondary conditions such as respiratory distress syndrome, which can result in higher health service use and costs, especially in the neonatal period. Children with multiple conditions would likely have higher health service use and costs, especially home health, due to their complex and special needs,

which has been found in previous studies (81). Unfortunately, effect modification of being born LBW or preterm on health care service use and costs among children with OFC could not be examined in this study due to sparse data.

In both groups of children, being born at a tertiary care center was positively associated with almost every health service use and cost category. These direct relationships may be due to tertiary care centers having more resources and medical technology to treat infants with special health care needs and children than community hospitals. Our results may also be due to children and families already in a referral system during the prenatal and/or postnatal period such as with receiving PNC and/or MCC. Cornerstones of MCC services are to develop a strong referral network, assist in accessing resources and ensure appropriate services are rendered, including continuity of care (162). Health care providers at tertiary care centers and community hospitals could be providing referrals and resources for inpatient and home health services. It also may be because most of the craniofacial centers and teams in North Carolina are affiliated with a tertiary care center (6, 124, 125). Hence, children with OFC being born at a tertiary care center could be directly referred to a craniofacial center or team at that hospital, which would explain why birth hospital would be associated with higher service use and cost, especially among this population. Having craniofacial centers and teams only in certain areas of the state may have also influenced geographical variations in service use and cost among children with OFC.

Low dental service use and cost among both groups of children was to be expected for several reasons. One, most children do not see a dentist during infancy (112, 163). In addition, previous studies have found unmet dental needs and low dental service use among children with and without special needs (95, 164, 165) and among poor and minority children

even when insurance was considered (131). Two, the results could likely be due to the shortage of Medicaid dental providers in North Carolina, especially the specialty needed to treat children with OFC (164, 166-168). Reasons for the low dentist participation rate in Medicaid include slow reimbursement, complicated paperwork, and the perception that Medicaid patients tend to miss appointments (167). Three, low service use and costs of general dental services for children with OFC may also be affected by parents not recognizing the need for dental care (165). Parents may place a higher priority on the more surgical, medical needs such as repairing the cleft rather than dental services children with OFC might need. Lastly, these results may be explained by the low or no reimbursement rates for some dental services by Medicaid (130, 132). Nevertheless, for children with OFC, it is important to see a dentist in the first year of life due to the need of specialized oral health services and higher risk of a variety of oral health conditions such as dental caries and periodontal disease (11, 112-114). This is a need that is clearly not being met and was recently recognized as such by the American Academy of Pediatric Dentistry (130).

This study also found underutilization of well-child care services among children with and without OFC. However, the extent of underutilization was greater among children with OFC. These results were consistent with previous studies on the low use of preventive services for children (169, 170). This low utilization has grave implications for both groups of children. Well-child care visits or the Early and Periodic Screening, Diagnostic, and Treatment (EPSDT) benefit is the package of Medicaid benefits for children and is called Health Check in North Carolina. These visits include comprehensive health and developmental assessments, hearing, vision, and dental services for children birth to 21 years of age. Well-child care visits can ultimately provide early identification of conditions that can impede

children's development (171-173). In addition, through EPSDT, children on Medicaid usually have more comprehensive coverage than a usual private health insurance plan, thereby increasing access to needed services to improve children's health and quality of life. Furthermore, CSHCN like children with OFC need more specialized services such as speech, physical, and occupational therapy, and mental health, which can be covered through the EPSDT benefits (171, 173). The low use of well-child care services may be due to restrictions on the frequency of such services in North Carolina and/or parents lack of knowledge such services are an entitlement for their children. In addition, it may be due to low provider participation in Medicaid as with dental care and/or inappropriate service denials due to managed care contracts (169, 173-175). Due to small numbers, possible individual and system characteristics associated with low well-child care utilization were unable to be examined in this study.

A reason for the low mental health and home health service use and cost among both groups of children may be due to the fact that parents may not see a need for such services and/or infants do not need these services, especially mental health services in the first year of life (81). During infancy, parents may focus on more urgent, medically and physically related needs such repairing the cleft among children with OFC and ear infections or chronic illness in children with and without OFC.

The results found in this study are similar to the few previous studies on health service utilization and costs of children with birth defects. Using Medicaid and hospital discharge datasets, Waitzman et al. found the per capita medical costs of infants with OFC in 1988 was $6,794, which was lower than in the present study. This is most likely due to inflation and the results being outdated (17). Using the consumer price index, this cost would amount to about

$11,625 today, which is still lower than what was found in this study. Two recent studies examining hospitalizations and costs in children with birth defects in the United States confirm the results found in this study. In 2003, Robbins et al. examined hospital costs during the newborn period from a sample of pediatric discharges in 36 states. Robbins et al. found that the mean hospital charge for children with CP alone was $33,387 and total hospital charges for children with CP alone were $72,914,132. For children with CL with or without CP, the mean hospital charge was $15,397 and total hospital charges were $53,630,046 (157). In a similar study, Russo et al. concluded that in 2004 the mean cost for children with CP alone was $5,400 and the aggregate costs were $15,506,700, and the mean cost for children with CL with or without CP was $5,500, which was similar to isolated CP. The aggregate costs for children with CL with or without CP were $27,155,800 (103). However, the unit of analyses in these studies was the hospital discharge and not the child and physician / professional fees were excluded. This probably explains why the present study has higher costs per child and lower overall costs for children with OFC than these previous studies.

Findings from this study are also similar to results of previous national and state studies on health care service utilization and costs among children in general, CSHCN, and medically fragile children (21, 66, 77, 95, 176, 177). Using the 1999 and 2000 Medical Expenditure Panel Survey data, Newacheck et al. found CSHCN had significantly higher hospitalizations, physician visits, and home health provider days than children without special needs and found that CSHCN had significantly higher health care expenditures than children without special needs (21, 95). However, dental service use was similar in children with and without special health care needs, as was found in the present study (95).

The results of this study are contrary to a study on health care costs of children born in 1997 and on Medicaid in North Carolina. Buescher et al. found that during the first year of life, child participation in WIC was associated with higher service use and Medicaid expenditures in physician, outpatient, prescription drug, and total Medicaid expenditures compared to children who did not participate in WIC (66). In the present study, WIC was inversely related to physician/ medical, outpatient, and total health service use and costs among children with and without OFC and only slightly associated with these outcomes among unaffected children.

Strengths and Potential Limitations

Potential limitations of this study include the study population and quality of the Medicaid and birth certificate data, which could have contributed to an underestimation of health service utilization and cost in both groups of children. Because this study restricted its analyses to children continuously enrolled in Medicaid, results were from only one type of health insurance and were not representative of all children in North Carolina. Utilization rates and reimbursement rates, which affect cost, can vary by health insurance type (81, 178). Moreover, children on Medicaid in other states could have different utilization rates and costs because reimbursement rates for Medicaid can vary from state-to-state, thereby limiting the generalizability of this study. Despite the limited generalizability, other states could use this study as a template to examine different categories of health service use and costs of other birth defects such as spina bifida and Down syndrome among children with public health insurance.

Restricting the analysis to children on Medicaid was also a strength in that children on Medicaid usually come from relatively low-income families, which made the study population more homogeneous and reduced the potential for confounding by socioeconomic status. It was also a strength because Medicaid paid claims are a bill paying system that require providers to report data for reimbursement; hence, there is an incentive for complete and accurate data (76, 145). Medicaid paid claims also included information on service use and costs outside of North Carolina, which provided a more comprehensive analysis of health service use and costs (176). By examining only those children continuously enrolled in Medicaid, a better depiction of service use and costs throughout the first year of life was obtained.

A variety of people can complete information on the birth certificate, such as mothers and nurses. This can lead to variability in data entry and influence validity and data quality. Maternal race/ethnicity can also be misclassified or underreported on the birth certificate (179), which could have affected the demographic and multivariate results for maternal race.

Another limitation was that certain important factors were unavailable for analysis in this study. These variables include: cleft laterality; occurrence of the same birth defect in families; mother and/or father's occupation; household income; previous participation or knowledge of services like MCC, WIC, Medicaid, and PNC; and frequency and duration of MCC and PNC services. Cleft laterality may have modified the effect of cleft type on service use and cost; however, the extent of this is unknown. Occurrence of the same birth defect in families of children with OFC and prior use of certain health services in both groups of children could potentially increase service use and thereby costs.

By excluding children that died within the first year of life, health care service use and costs may have been underestimated as those children probably utilized more health services and had higher costs due to their severe and complex medical conditions. Exclusion of these children would under-estimate true health service utilization and costs in both groups of children. However, these children would not have met the definition of continuous enrollment and as such should have been excluded from the analysis.

This study only included direct health care costs, which is yet another limitation. Other costs for children with OFC include caregiver and psychosocial costs, mortality, morbidity, and disability costs, and out-of-pocket costs for equipment and appliances not covered by Medicaid such as some pre- and post-orthodontic appliances. Hence, this study represents only a portion of total costs for children with OFC. This study also excluded prescription medication usage and costs because data were unavailable, which can lead to an underestimation of service use and costs in both groups of children.

Another potential limitation is that some results such as "Other" race/ethnicity and "noncore areas" with place of residence should be interpreted with caution. Population sample sizes for mental and home health service use and cost along with small cell sizes for "Other" race and some categories of place of residence had wide confidence intervals, which lead to imprecise effect estimates, especially among children with OFC.

Despite these limitations, this study has several strengths because employing birth defects data and covariates have not been previously examined in health service use and cost studies. This study employed a population-based birth defects surveillance system to verify the condition whereas previous studies using Medicaid diagnoses codes did not. As a result, service use and cost by cleft type and presence of other anomalies were examined. The few

previous studies on costs of children with birth defects that have included OFC have only analyzed CL with or without CP and CL alone, which masks the effects of CP alone. Additionally, these studies did not examine the presence of other anomalies, which affects service use and cost as clearly indicated in this study.

This study is the first to examine the effect of certain variables such as cleft type, presence of other anomalies, LBW, preterm birth, PNC, and MCC have on health care service utilization and costs. It is important to evaluate service use and costs by these variables because service use and costs patterns for different services may vary depending on the condition examined. If a child has multiple conditions, such as LBW, CLP, and another birth defect, this would increase service use and costs compared to a child that just had CL alone.

Another strength is that this study examined the effect of geographic variables like perinatal care region and place of residence have on service utilization and costs, which have not been previously studied. Some Title V services, including genetics, still use perinatal care regions of residence to organize service delivery of care within the state (148, 149). The Urban Influence Codes were employed to examine how adjacency to metropolitan areas affects costs. This method is beneficial when examining the structure of health care systems and whether outcomes or care are possibly related to the complexity of the medical or health community (150, 151). This typology is helpful in distinguishing the effects of adjacency to a metropolitan area or small town compared to the size of the area on access and health service utilization and costs (152). Including these geographic variables can assist in targeting resources and services to populations in need of services such as in the southeastern and western perinatal care regions and in noncore areas adjacent and not adjacent to metropolitan

areas. Most previous studies on health service use and cost of CSHCN used traditional urban and rural classifications, which do not consider adjacency to urban areas.

Other strengths of this study included statewide information on health services and costs by specific health categories for children with and without OFC. Previous studies on health service use and cost of children with OFC were not as comprehensive and did not include a comparison group. In this study the unit of analysis was a child compared to previous health service use and cost studies on birth defects where the unit of analysis was the hospital discharge or visit. This study examined actual costs reimbursed by Medicaid and not charges made to better understand the direct financial burden of families affected by OFC.

Public Health Implications

Understanding the nature of health care service utilization and costs among children with OFC are important for health planning efforts and delivery of services by public agencies such as federal, state, and local Title V CSHCN programs (95). This study indicates that certain populations like less educated, younger and older mothers, minority populations, and families living in certain areas of the state are less likely to use specific health services such as home health and mental health. This is probably associated with lack of access to such services and/or lack of awareness or knowledge these families are eligible for such services (180-182). This study also underscores the importance for resources and trained health care providers, especially in home and mental health and dental services, for families of children with OFC. This study also indicates that receipt of certain services such as initiation of PNC in the first trimester, WIC, and MCC can be positively or negatively associated with outpatient, mental health, home health, and total service use and costs in children with and

without OFC. This is important because maternity outreach workers, community transition coordinators and case managers can utilize this information to link families and children to services and ensure a referral system is in place for children, in particular, CSHCN such as children with OFC. This information can also be used to address the research priorities for children with OFC as recently identified by CDC (33).

In general, children with OFC and unaffected children tended to differ in maternal, child, and system characteristics that were associated with less total service use and cost. As such, populations that need to be targeted will differ for cases and controls, which will affect service delivery and cost initiatives. For example, children without OFC may not need as many inpatient, well-child care, and home health services covered under Medicaid whereas children with OFC may need increased Medicaid coverage with inpatient, well-child care, and home health services to receive specialized services for their condition.

This study provides information on patterns and effects of individual and system characteristics on health care service utilization and costs that may be applied in determining appropriate benefit packages and efficient payment mechanisms for both private and public health insurance systems among children with and without OFC (95). Specifically, this information can be used to identify: populations in need of services; availability of services; and categories of high and low utilization and cost. Such information can be used by the North Carolina Commission on Children with Special Health Care Needs, which makes recommendations for modifications or additions to rules regarding service provision and delivery among CSHCN in the state.

Differences in service use and cost by maternal race/ethnicity, age, or education, perinatal care region or place of residence among children with OFC should be considered in

organizing service needs and evaluating pediatric initiatives at the state level (183). This is because this population is at risk for learning disabilities and developing dental caries as well as other secondary conditions due to their OFC. If the health needs of this population are not addressed, these children could face poor health outcomes when OFC is a repairable birth defect.

Another public health implication is that differences in service use and costs, especially among children with OFC, illustrate the importance of developing systems of coordinated care like medical homes to manage complex chronic conditions, such as children with birth defects, in low-income populations as indicated by the American Academy of Pediatrics and the National Center of Medical Home Initiatives for Children with Special Health Care Needs (8, 115, 116). This is especially important for children with multiple anomalies who tend to utilize significantly more health services and cost significantly more than children with isolated OFC as shown in this study. Children with multiple birth defects need coordinated care to help manage their unique, complex conditions. Determining factors that mediate Medicaid costs will guide future investments in public programs that coordinate care or otherwise serve families of children affected by OFC. In fact, previous research has shown that care coordination can improve health outcomes and reduce costs (184-186). Furthermore, it is well known that early intervention has an important positive impact on CSHCN (187-190), and studies such as this one can help identify and refer certain families of children with OFC for early intervention.

Lastly, this study also underscores the importance of continued health insurance and adequate coverage for CSHCN such as children with birth defects to have access to services needed to improve the overall health of these children. All of these issues are particularly

important as many states are facing budget deficits and concern that states may reduce the coverage provided or individuals enrolled in the Medicaid program. Reducing coverage may escalate national and state health care spending, not reduce it. Increased health care costs would then be due to greater utilization of emergency services, decreased availability of specialized services and preventive services, and increased numbers of underinsured and uninsured children.

Conclusion

This chapter determines how maternal, child, and system characteristics affect health service use and cost among children with and without OFC during the first year of life. Such information can lead to targeting populations which are experiencing unmet service needs and improve service availability and delivery in certain areas of the state. This chapter also provides unprecedented data on health service use and cost of children with OFC by cleft type and presence of other anomalies, which can assist in targeting care coordination and early intervention. Results from this study provide accurate and current data for service planning, program planning, and policy development. This information is a first step toward ensuring and increasing access to services and ultimately improving the overall health and development of children with OFC and other birth defects in North Carolina.

Table 2.1. Selected characteristics of children with and without orofacial clefts during the first year of life in North Carolina, 1995-2002[a]

Characteristic	Children with Orofacial Clefts N = 565 (%)	Children without Orofacial Clefts N = 5,674 (%)	P-value
Maternal			
Age			
≤ 20 years old	173 (30.6)	1,960 (34.5)	0.07
21-24 years old	166 (29.4)	1,705 (30.1)	
25-29 years old	141 (25.0)	1,171 (20.6)	
≥ 30 years old	85 (15.0)	838 (14.8)	
Education			
< High school	247 (43.7)	2,339 (41.2)	0.51
High school	213 (37.7)	2,224 (39.2)	
> High school	105 (18.6)	1,111 (19.6)	
Race			
White/non-Hispanic	348 (61.6)	2,633 (46.4)	<0.00*
Black/non-Hispanic	126 (22.3)	2,078 (36.6)	
Hispanic	69 (12.2)	755 (13.3)	
Other[b]	22 (3.9)	208 (3.7)	
Number of Living Children			
0	249 (44.1)	2,493 (44.0)	0.64
1	165 (29.2)	1,749 (30.8)	
≥ 2	151 (26.7)	1,431 (25.2)	
Marital Status			
Married	259 (45.8)	2,313 (40.8)	0.02*
Not married	306 (54.2)	3,361 (59.2)	
Initiation of Prenatal Care in First Trimester			
Yes	435 (77.0)	4,239 (74.7)	0.40
No	128 (22.7)	1,400 (24.7)	
Child			
Birthweight			
< 2,500 grams	82 (14.5)	505 (8.9)	<0.00*
≥ 2,500 grams	483 (85.5)	5,169 (91.1)	
Preterm Birth			
< 37 weeks	85 (15.0)	628 (11.1)	0.00*
≥ 37 weeks	480 (85.0)	5,046 (88.9)	
Gender			
Female	248 (43.9)	2,759 (48.6)	0.00*
Male	317 (56.1)	2,915 (51.4)	
Cleft Type			
Cleft lip only	108 (19.1)	n/a	n/a
Cleft palate only	210 (37.2)	n/a	
Cleft lip with cleft palate	247 (43.7)	n/a	

Characteristic	Children with Orofacial Clefts N = 565 (%)	Children without Orofacial Clefts N = 5,674 (%)	P-value
Child (cont.)			
Presence of Other Anomalies[c]			
Isolated	368 (65.1)	n/a	n/a
Multiple	197 (34.9)	n/a	
System			
Source of Prenatal Care			
Health department	350 (62.0)	3,341 (58.9)	0.16
Other	215 (38.1)	2,333 (41.1)	
Receipt of Maternity Care Coordination Services			
Yes	252 (44.6)	2,573 (45.4)	0.73
No	313 (55.4)	3,101 (54.7)	
Receipt of WIC			
Yes	387 (68.5)	4,129 (72.8)	0.03*
No	178 (31.5)	1,545 (27.2)	
Birth Hospital Level of Care			
Level III	241 (42.7)	2,184 (38.5)	0.05
Community	324 (57.4)	3,490 (61.5)	
Perinatal Care Region			
Northwestern	143 (25.3)	1,374 (24.2)	0.08
Southwestern	96 (17.0)	808 (14.2)	
Northeastern	92 (16.3)	926 (16.3)	
Southeastern	74 (13.1)	974 (17.2)	
Eastern	99 (17.5)	1,068 (18.8)	
Western	61 (10.8)	524 (9.2)	
Place of Residence			
Metropolitan	357 (63.2)	3,514 (61.9)	0.90
Micropolitan	134 (23.7)	1,374 (24.2)	
Noncore areas adjacent to metro area or small town	52 (9.2)	532 (9.4)	
Noncore areas not adjacent to metro area or small town	22 (3.9)	254 (4.5)	

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly = orofacial cleft diagnosis only; multiple anomalies = orofacial cleft + other birth defect diagnosis
* $P < 0.05$

Table 2.2. Mean number of paid claims per child and range among children with and without orofacial clefts during the first year of life in North Carolina, 1995-2002[a]

Service Category	Children with Orofacial Clefts (n = 565)		Children without Orofacial Clefts (n = 5,674)		Utilization Ratio[b]
	Mean	Range	Mean	Range	
Medical	60.12	2.00-491.00	36.75	0.00-258.00	1.64*
Inpatient	1.95	1.00-12.00	1.19	0.00-10.00	1.64*
Outpatient	5.12	0.00-44.00	1.92	0.00-33.00	2.67*
Mental Health	1.98	0.00-56.00	0.10	0.00-20.00	19.80*
Home Health	1.43	0.00-68.00	0.08	0.00-36.00	17.88*
Dental	0.18	0.00-5.00	0.00	0.00-1.00	0.00*
Well-Child Care	3.70	0.00-9.00	3.83	0.00-11.00	0.97
Other	0.21	0.00-10.00	0.05	0.00-6.00	4.20*
Total	74.70	6.00-530.00	43.92	1.00-315.00	1.70*

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Utilization ratio = ratio of mean number of paid claims per child with orofacial clefts to mean number of paid claims per child without orofacial clefts
* Utilization ratios for all categories of service except well-child care were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 2.3. Mean number of paid claims per child and range among children with orofacial clefts by cleft type during the first year of life in North Carolina, 1995-2002[a]

Service Category	Cleft Type					
	CL[b] (n = 108)		CP[b] (n = 210)		CLP[b] (n = 247)	
	Mean	Range	Mean	Range	Mean	Range
Medical	45.23	7.00-137.00	62.73	2.00-391.00	64.41	9.00-491.00
Inpatient	1.34	1.00-3.00	2.02	1.00-10.00	2.16	1.00-12.00
Outpatient	3.27	0.00-17.00	5.13	0.00-35.00	5.92	0.00-44.00
Mental Health	0.49	0.00-15.00	2.11	0.00-56.00	2.53	0.00-42.00
Home Health	0.08	0.00-3.00	1.74	0.00-68.00	1.77	0.00-52.00
Dental	0.13	0.00-2.00	0.16	0.00-3.00	0.21	0.00-5.00
Well-Child Care	3.98	0.00-8.00	3.66	0.00-9.00	3.62	0.00-8.00
Other	0.01	0.00-1.00	0.28	0.00-10.00	0.24	0.00-4.00
Total	54.54	12.00-156.00	77.83	6.00-414.00	80.85	12.00-530.00

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] CL = cleft lip; CP = cleft palate; CLP = cleft lip with cleft palate

Table 2.4. Mean number of paid claims per child and range among children with orofacial clefts by presence of other anomalies during the first year of life in North Carolina, 1995-2002[a]

Service Category	Presence of Other Anomalies[b]			
	Isolated Anomaly (n=368)		Multiple Anomalies (n=197)	
	Mean	Range	Mean	Range
Medical	48.43	7.00-134.00	81.95	2.00-491.00
Inpatient	1.60	1.00-6.00	2.61	1.00-12.00
Outpatient	3.95	0.00-19.00	7.30	0.00-44.00
Mental Health	1.32	0.00-56.00	3.22	0.00-42.00
Home Health	0.31	0.00-44.00	3.54	0.00-14.00
Dental	0.17	0.00-5.00	0.19	0.00-3.00
Well-Child Care	3.87	0.00-8.00	3.39	0.00-9.00
Other	0.06	0.00-2.00	0.49	0.00-10.00
Total	59.71	12.00-159.00	102.69	6.00-530.00

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Isolated anomaly = orofacial cleft diagnosis only; multiple anomalies = orofacial cleft + other birth defect diagnosis

Table 2.5. Mean cost and range in dollars per child with and without orofacial clefts during the first year of life in North Carolina, 1995-2002[a]

Cost Category	Children with Orofacial Clefts (n = 565)		Children without Orofacial Clefts (n = 5,674)		Cost Ratio[b]
	Mean	Range	Mean	Range	
Medical	4,759	74-152,898	1,174	0-40,854	4.05*
Inpatient	11,591	0-598,919	2,149	0-268,948	5.39*
Outpatient	3,482	0-19,526	221	0-9,915	15.76*
Mental Health	632	0-61,169	17	0-4,395	37.18*
Home Health	1,843	0-133,990	41	0-93,846	44.95*
Dental	13	0-1,479	0	0-72	0.00*
Well-Child Care	277	0-714	290	0-796	0.96
Other	44	0-2,070	8	0-2026	5.50*
Total	22,642	74-680,374	3,900	0-404,760	5.81*

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Cost ratio = ratio of mean cost per child with orofacial clefts to mean cost per child without orofacial clefts
* Cost ratios for all cost categories except well-child care were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 2.6. Mean cost and range in dollars per child with orofacial clefts by cleft type during the first year of life in North Carolina, 1995-2002[a]

Service Category	Cleft Type					
	CL[b] (n = 108)		CP[b] (n = 210)		CLP[b] (n = 247)	
	Mean	Range	Mean	Range	Mean	Range
Medical	2,351	180-11,635	5,248	74-152,898	5,396	149-65,738
Inpatient	2,361	0-28,756	14,312	0-598,919	13,314	0-436,997
Outpatient	2,823	0-9,175	2,044	0-19,526	4,992	0-16,907
Mental Health	79	0-2,653	995	0-61,169	566	0-11,638
Home Health	46	0-2,214	2,324	0-133,990	2,220	0-116,273
Dental	5	0-102	11	0-830	19	0-1,479
Well-Child Care	300	0-554	274	0-714	270	0-627
Other	9	0-937	68	0-2,070	40	0-1,583
Total	7,973	1,101-46,739	25,575	74-680,374	26,817	465-504,374

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] CL = cleft lip; CP = cleft palate; CLP = cleft lip with cleft palate

Table 2.7. Mean cost and range in dollars per child with orofacial clefts by presence of other anomalies during the first year of life in North Carolina, 1995-2002[a]

Service Category	Presence of Other Anomalies[b]			
	Isolated Anomaly		Multiple Anomalies	
	Mean	Range	Mean	Range
Medical	2,790	149-17,225	8,436	74-152,898
Inpatient	3,097	0-36,323	27,459	0-598,919
Outpatient	3,511	0-17,150	3,426	0-19,526
Mental Health	247	0-7,269	1,353	0-61,169
Home Health	140	0-12,157	5,025	0-133,990
Dental	14	0-1,479	13	0-891
Well-Child Care	290	0-627	253	0-714
Other	11	0-937	107	0-2,070
Total	10,099	465-55,646	46,072	74-680,374

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Isolated anomaly = orofacial cleft diagnosis only; multiple anomalies = orofacial cleft + other birth defect diagnosis

Table 2.8. Binary logit regression of selected characteristics on *any* outpatient health care costs among children with and without orofacial clefts during the first year of life in North Carolina, 1995-2002[a]

Characteristics	Children with Orofacial Clefts (n=563)		Children without Orofacial Clefts (n=5,638)	
	Odds Ratio[†]	95% Confidence Intervals	Odds Ratio[†]	9 5% Confidence Intervals
Maternal				
Age				
≤ 20 years old	0.71	0.26, 1.95	1.01	0.87, 1.18
21-24 years old	1.00		1.00	
25-29 years old	2.43	0.88, 6.72	0.77	0.66, 0.91*
≥ 30 years old	2.06	0.62, 6.81	0.83	0.69, 0.99*
Education				
< High school	4.15	1.35, 12.75*	1.36	1.15, 1.61*
High school	1.02	0.40, 2.60	1.13	0.97, 1.32
> High school	1.00		1.00	
Race				
White/non-Hispanic	1.00		1.00	
Black/non-Hispanic	0.62	0.24, 1.63	1.23	1.07, 1.42*
Hispanic	0.92	0.26, 3.29	1.25	1.03, 1.52*
Other[b]	**		0.85	0.63, 1.14
Number of Living Children				
0	1.00		1.00	
1	0.29	0.16, 0.72*	0.87	0.75, 1.00
≥ 2	0.40	0.13, 1.25	0.83	0.70, 0.98*
Marital Status				
Married	1.00		1.00	
Not married	0.88	0.38, 2.01	1.09	0.96, 1.23
Initiation of Prenatal Care in First Trimester				
Yes	1.00		1.00	
No	1.26	0.48, 3.34	0.83	0.73, 0.95*
Child				
Birthweight				
< 2,500 grams	**		1.22	0.94, 1.58
≥ 2,500 grams	1.00		1.00	
Preterm Birth				
< 37 weeks	**		1.46	1.15, 1.85*
≥ 37 weeks	1.00		1.00	
Gender				
Female	1.00		1.00	
Male	1.00	0.49, 2.04	1.35	1.20, 1.50*
System				
Source of Prenatal Care				
Health department	1.00		1.00	
Other	1.39	0.61, 3.21	0.99	0.88, 1.13
Receipt of Maternity Care Coordination Services				
Yes	1.64	0.68, 3.92	1.05	0.92, 1.20
No	1.00		1.00	

Characteristics	Children with Orofacial Clefts (n=563)		Children without Orofacial Clefts (n=5,638)	
	Odds Ratio[†]	95% Confidence Intervals	Odds Ratio[†]	9 5% Confidence Intervals
System (cont.)				
Receipt of WIC				
Yes	0.42	0.16, 1.11	1.16	1.01, 1.33[*]
No	1.00		1.00	
Birth Hospital Level of Care				
Level III	0.87	0.36, 2.11	1.10	0.96, 1.26
Community	1.00		1.00	
Perinatal Care Region				
Northwestern	1.00		1.00	
Southwestern	0.76	0.26, 2.17	1.18	0.98, 1.42
Northeastern	1.23	0.38, 4.03	1.11	0.92, 1.34
Southeastern	2.79	0.63, 12.32	1.17	0.97, 1.41
Eastern	1.22	0.41, 3.64	0.87	0.73, 1.05
Western	1.34	0.32, 5.57	1.04	0.84, 1.30
Place of Residence				
Metropolitan	1.00		1.00	
Micropolitan	0.92	0.36, 2.33	1.11	0.96, 1.30
Noncore adjacent	0.47	0.13, 1.63	1.32	1.07, 1.64[*]
Noncore areas not adjacent	**		1.24	0.92, 1.66

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
** Unable to determine estimate due to insufficient cell sizes
[†] Adjusted for all covariates in model

Table 2.9. Binary logit regression of selected characteristics on *any* mental health care costs among children with and without orofacial clefts during the first year of life in North Carolina, 1995-2002[a]

Characteristics	Children with Orofacial Clefts (n=563)		Children without Orofacial Clefts (n=5,638)	
	Odds Ratio[†]	95% Confidence Intervals	Odds Ratio[†]	95% Confidence Intervals
Maternal				
Age				
≤ 20 years old	1.01	0.55, 1.89	0.67	0.36, 1.25
21-24 years old	1.00		1.00	
25-29 years old	0.99	0.54, 1.78	1.03	0.55, 1.91
≥ 30 years old	1.26	0.62, 2.54	0.95	0.47, 1.90
Education				
< High school	1.25	0.63, 2.49	1.31	0.67, 2.54
High school	1.18	0.61, 2.28	0.85	0.45, 1.64
> High school	1.00		1.00	
Race				
White/non-Hispanic	1.00		1.00	
Black/non-Hispanic	0.76	0.40, 1.42	1.04	0.59, 1.84
Hispanic	1.51	0.75, 3.03	1.22	0.57, 2.58
Other[b]	2.44	0.84, 7.06	0.64	0.15, 2.76
Number of Living Children				
0	1.00		1.00	
1	1.77	1.04, 3.02*	1.02	0.58, 1.80
≥ 2	0.98	0.52, 1.86	1.17	0.62, 2.19
Marital Status				
Married	1.00		1.00	
Not married	1.00	0.61, 1.62	1.28	0.76, 2.15
Initiation of Prenatal Care in First Trimester				
Yes	1.00		1.00	
No	1.65	0.98, 2.78	1.53	0.94, 2.48
Child				
Birthweight				
< 2,500 grams	1.56	0.75, 3.23	4.04	2.11, 7.75*
≥ 2,500 grams	1.00		1.00	
Preterm Birth				
< 37 weeks	2.15	1.03, 4.48*	2.40	1.24, 4.63*
≥ 37 weeks	1.00		1.00	
Gender				
Female	1.00		1.00	
Male	1.10	0.70, 1.71	1.74	1.11, 2.75
System				
Source of Prenatal Care				
Health department	1.00		1.00	
Other	1.53	0.91, 2.58	0.88	0.54, 1.43
Receipt of Maternity Care Coordination Services				
Yes	1.28	0.77, 2.12	1.87	1.11, 3.15*
No	1.00		1.00	

Characteristics	Children with Orofacial Clefts (n=563) Odds Ratio[†]	95% Confidence Intervals	Children without Orofacial Clefts (n=5,638) Odds Ratio[†]	95% Confidence Intervals
System (cont.)				
Receipt of WIC				
Yes	1.27	0.75, 2.17	0.54	0.32, 0.91[*]
No	1.00		1.00	
Birth Hospital Level of Care				
Level III	1.09	0.65, 1.82	0.94	0.56, 1.60
Community	1.00		1.00	
Perinatal Care Region				
Northwestern	1.00		1.00	
Southwestern	0.97	0.51, 1.85	0.52	0.23, 1.17
Northeastern	0.51	0.24, 1.05	0.43	0.20, 0.94[*]
Southeastern	0.44	0.19, 1.02	0.42	0.19, 0.93[*]
Eastern	0.54	0.26, 1.12	0.55	0.26, 1.15
Western	0.77	0.34, 1.72	1.72	0.88, 3.33
Place of Residence				
Metropolitan	1.00		1.00	
Micropolitan	1.05	0.58, 1.89	1.24	0.70, 2.20
Noncore adjacent	3.15	1.48, 6.74[*]	1.65	0.80, 3.38
Noncore areas not adjacent	3.95	1.32, 11.83[*]	1.88	0.70, 5.05

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
** Unable to determine estimate due to insufficient cell sizes
[†] Adjusted for all covariates in model

Table 2.10. Binary logit regression of selected characteristics on *any* home health care costs among children with and without orofacial clefts during the first year of life in North Carolina, 1995-2002[a]

Characteristics	Children with Orofacial Clefts (n=563)		Children without Orofacial Clefts (n=5,638)	
	Odds Ratio[†]	95% Confidence Intervals	Odds Ratio[†]	95% Confidence Intervals
Maternal				
Age				
≤ 20 years old	0.98	0.52, 1.86	0.72	0.45, 1.14
21-24 years old	1.00		1.00	
25-29 years old	1.15	0.62, 2.14	1.06	0.65, 1.74
≥ 30 years old	0.67	0.29, 1.54	1.34	0.79, 2.26
Education				
< High school	1.05	0.51, 2.16	1.61	0.96, 2.69
High school	1.02	0.52, 1.99	1.05	0.64, 1.72
> High school	1.00		1.00	
Race				
White/non-Hispanic	1.00		1.00	
Black/non-Hispanic	0.83	0.43, 1.59	0.74	0.49, 1.13
Hispanic	0.71	0.31, 1.59	0.62	0.34, 1.14
Other[b]	0.93	0.30, 2.88	0.79	0.32, 1.92
Number of Living Children				
0	1.00		1.00	
1	1.11	0.64, 1.92	0.91	0.61, 1.36
≥ 2	0.61	0.31, 1.20	0.53	0.31, 0.88[*]
Marital Status				
Married	1.00		1.00	
Not married	0.91	0.54, 1.53	1.17	0.79, 1.73
Initiation of Prenatal Care in First Trimester				
Yes	1.00		1.00	
No	0.85	0.45, 1.53	1.09	0.73, 1.63
Child				
Birthweight				
< 2,500 grams	2.83	1.31, 6.11[*]	2.31	1.39, 3.83[*]
≥ 2,500 grams	1.00		1.00	
Preterm Birth				
< 37 weeks	0.92	0.41, 2.08	3.43	2.10, 5.59[*]
≥ 37 weeks	1.00		1.00	
Gender				
Female	1.00		1.00	
Male	1.28	0.79, 2.06	1.85	1.31, 2.62[*]
System				
Source of Prenatal Care				
Health department	1.00		1.00	
Other	1.06	0.61, 1.83	0.95	0.65, 1.37
Receipt of Maternity Care Coordination Services				
Yes	1.75	1.00, 3.04[*]	1.05	0.71, 1.55
No	1.00		1.00	

Characteristics	Children with Orofacial Clefts (n=563) Odds Ratio[†]	95% Confidence Intervals	Children without Orofacial Clefts (n=5,638) Odds Ratio[†]	95% Confidence Intervals
System (cont.)				
Receipt of WIC				
Yes	0.53	0.30, 0.93[*]	0.99	0.66, 1.47
No	1.00		1.00	
Birth Hospital Level of Care				
Level III	1.81	1.05, 3.11[*]	1.97	1.3, 2.91[*]
Community	1.00		1.00	
Perinatal Care Region				
Northwestern	1.00		1.00	
Southwestern	2.04	0.96, 4.32	1.00	0.61, 1.64
Northeastern	1.32	0.59, 2.96	0.19	0.09, 0.40[*]
Southeastern	3.41	1.50, 7.75[*]	0.92	0.57, 1.49
Eastern	1.72	0.77, 3.84	0.40	0.21, 0.79[*]
Western	1.97	0.84, 4.65	0.97	0.55, 1.74
Place of Residence				
Metropolitan	1.00		1.00	
Micropolitan	1.21	0.65, 2.26	0.94	0.59, 1.49
Noncore adjacent	1.18	0.51, 2.74	0.64	0.30, 1.33
Noncore areas not adjacent	1.16	0.29, 4.71	0.91	0.30, 2.79

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
** Unable to determine estimate due to insufficient cell sizes
[†] Adjusted for all covariates in the model

Table 2.11. Ordinary least squares regression of selected characteristics on medical costs among children with and without orofacial clefts with medical costs during the first year of life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Cost Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=5,633)** Cost Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	0.90 (0.72, 1.12)	0.99 (0.94, 1.05)
21-24 years old	1.00	1.00
25-29 years old	0.87 (0.70, 1.08)	0.97 (0.91, 1.03)
≥ 30 years old	0.73 (0.57, 0.95)*	1.01 (0.95, 1.08)
Education		
< High school	1.19 (0.94, 1.52)	1.11 (1.04, 1.18)*
High school	1.01 (0.80, 1.26)	1.03 (0.97, 1.09)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	0.77 (0.62, 0.95)*	0.96 (0.92, 1.01)
Hispanic	0.86 (0.66, 1.11)	0.89 (0.83, 0.95)*
Other[b]	1.20 (0.79, 1.81)	0.96 (0.86, 1.07)
Number of Living Children		
0	1.00	1.00
1	1.00 (0.83, 1.22)	0.94 (0.89, 0.99)*
≥ 2	1.05 (0.84, 1.30)	0.90 (0.85, 0.96)*
Marital Status		
Married	1.00	1.00
Not married	0.92 (0.77, 1.09)	1.07 (1.02, 1.12)*
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	1.05 (0.86, 1.27)	0.89 (0.85, 0.93)*
Child		
Birthweight		
< 2,500 grams	1.34 (1.00, 1.80)	1.61 (1.47, 1.76)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.45 (1.08, 1.95)*	1.47 (1.35, 1.59)*
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	1.11 (0.95, 1.30)	1.26 (1.21, 1.31)*

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.89 (0.74, 1.07)	0.96 (0.92, 1.01)
Receipt of Maternity Care Coordination Services		
Yes	1.24 (1.03, 1.49)[*]	1.10 (1.05, 1.16)[*]
No	1.00	1.00
Receipt of WIC		
Yes	0.87 (0.72, 1.06)	1.07 (1.02, 1.12)[*]
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.26 (1.05, 1.52)[*]	1.01 (0.96, 1.06)
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	1.03 (0.80, 1.31)	1.18 (1.10, 1.26)[*]
Northeastern	1.04 (0.81, 1.34)	0.99 (0.93, 1.06)
Southeastern	1.26 (0.95, 1.67)	1.07 (1.01, 1.15)[*]
Eastern	1.04 (0.80, 1.34)	1.00 (0.94, 1.07)
Western	1.11 (0.83, 1.50)	1.24 (1.15, 1.35)[*]
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	0.99 (0.80, 1.22)	1.02 (0.97, 1.08)
Noncore adjacent	0.83 (0.62, 1.11)	1.03 (0.96, 1.11)
Noncore areas not adjacent	1.29 (0.83, 2.01)	1.12 (1.01. 1.25)[*]

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 2.12. Ordinary least squares regression of selected characteristics on inpatient costs among children with and without orofacial clefts with inpatient costs during the first year of life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=550)** Cost Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=5,552)** Cost Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	1.03 (0.76, 1.41)	0.98 (0.93, 1.04)
21-24 years old	1.00	1.00
25-29 years old	1.06 (0.78, 1.43)	0.98 (0.92, 1.04)
≥ 30 years old	0.70 (0.49, 1.02)	0.94 (0.88, 1.01)
Education		
< High school	1.12 (0.80, 1.58)	1.07 (1.01, 1.14)[*]
High school	1.05 (0.76, 1.45)	1.01 (0.96, 1.07)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	0.80 (0.59, 1.09)	1.00 (0.95, 1.05)
Hispanic	0.82 (0.56, 1.19)	0.96 (0.89, 1.03)
Other[b]	1.12 (0.63, 2.02)	1.01 (0.91, 1.13)
Number of Living Children		
0	1.00	1.00
1	1.15 (0.87, 1.52)	0.99 (0.94, 1.04)
≥ 2	1.26 (0.91, 1.72)	0.99 (0.93, 1.05)
Marital Status		
Married	1.00	1.00
Not married	0.84 (0.65, 1.08)	1.03 (0.99, 1.08)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	0.98 (0.75, 1.29)	0.98 (0.93, 1.03)
Child		
Birthweight		
< 2,500 grams	1.96 (1.29, 2.98)[*]	2.52 (2.30. 2.76)[*]
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.77 (1.16, 2.69)[*]	2.01 (1.85, 2.18)[*]
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	0.93 (0.74, 1.16)	1.12 (1.07, 1.16)[*]

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.90 (0.70, 1.18)	0.97 (0.92, 1.01)
Receipt of Maternity Care Coordination Services		
Yes	1.35 (1.04, 1.75)[*]	1.03 (0.98, 1.08)
No	1.00	1.00
Receipt of WIC		
Yes	0.91 (0.69, 1.19)	1.00 (0.95, 1.05)
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.60 (1.23, 2.08)[*]	1.29 (1.23, 1.36)[*]
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	1.05 (0.74, 1.49)	1.06 (0.99, 1.13)
Northeastern	1.04 (0.73, 1.50)	1.11 (1.04, 1.19)[*]
Southeastern	1.25 (0.84, 1.88)	1.06 (0.99, 1.13)
Eastern	1.16 (0.81, 1.67)	1.09 (1.02, 1.16)[*]
Western	1.18 (0.78, 1.80)	1.01 (0.93, 1.10)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	1.01 (0.75, 1.35)	1.01 (0.95, 1.06)
Noncore adjacent	0.95 (0.62, 1.45)	1.02 (0.94, 1.10)
Noncore areas not adjacent	1.34 (0.72, 2.49)	1.00 (0.90, 1.12)

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 2.13. Ordinary least squares regression of selected characteristics on outpatient costs among children with and without orofacial clefts with outpatient costs during the first year of life in North Carolina, 1995-2002[a]

Characteristics	Children with Orofacial Clefts (n=513) Cost Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts (n=3,611) Cost Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	0.69 (0.45, 1.05)	0.94 (0.83, 1.06)
21-24 years old	1.00	1.00
25-29 years old	0.51 (0.34, 0.78)*	0.99 (0.87, 1.13)
≥ 30 years old	0.70 (0.43,1.15)	0.95 (0.82, 1.11)
Education		
< High school	1.56 (0.99, 2.48)	1.15 (1.00, 1.32)*
High school	1.19 (0.77, 1.84)	1.08 (0.95, 1.23)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	0.91 (0.60, 1.39)	1.11 (0.99, 1.24)
Hispanic	0.99 (0.61, 1.62)	1.08 (0.93, 1.25)
Other[b]	1.42 (0.66, 3.05)	1.32 (1.03, 1.71)*
Number of Living Children		
0	1.00	1.00
1	0.91 (0.62, 1.32)	0.98 (0.87, 1.09)
≥ 2	0.86 (0.56, 1.31)	1.06 (0.93, 1.21)
Marital Status		
Married	1.00	1.00
Not married	1.09 (0.78, 1.53)	1.05 (0.95, 1.16)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	1.13 (0.79, 1.63)	0.84 (0.76, 0.93)*
Child		
Birthweight		
< 2,500 grams	1.06 (0.62, 1.82)	1.32 (1.10, 1.58)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.22 (0.71, 2.10)	1.12 (0.95, 1.33)
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	1.45 (1.07, 1.96)*	1.26 (1.16, 1.38)*

Characteristics	Children with Orofacial Clefts Cost Ratios† (95% Confidence Intervals)	Children without Orofacial Clefts Cost Ratios† (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.83 (0.58, 1.18)	1.11 (1.01, 1.22)*
Receipt of Maternity Care Coordination Services		
Yes	0.87 (0,53, 1.09)	1.05 (0.94, 1.16)
No	1.00	1.00
Receipt of WIC		
Yes	0.89 (0.62, 1.27)	1.13 (1.01, 1.26)*
No	1.00	1.00
Birth Hospital Level of Care		
Level III	0.76 (0.53, 1.09)	1.20 (1.08, 1.34)*
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	0.87 (0.54, 1.40)	0.97 (0.84. 1.13)
Northeastern	1.22 (0.75, 1.98)	1.37 (1.18, 1.59)*
Southeastern	1.12 (0.65, 1.90)	0.90 (0.78, 1.04)
Eastern	0.48 (0.29, 0.78)	0.84 (0.72, 0.97)*
Western	0.24 (0.13, 0.42)*	1.05 (0.88, 1.25)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	0.84 (0.57, 1.26)	0.88 (0.78, 0.99)*
Noncore adjacent	0.57 (0.32, 1.01)	1.08 (0.92, 1.28)
Noncore areas not adjacent	0.67 (0.29, 1.51)	1.05 (0.83, 1.34)

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
* Statistically significant
† Adjusted for all covariates in the model

Table 2.14. Ordinary least squares regression of selected characteristics on mental health costs among children with and without orofacial clefts with mental health costs during the first year of life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=121)** Cost Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=87)** Cost Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	0.74 (0.35, 1.56)	0.81 (0.32, 2.06)
21-24 years old	1.00	1.00
25-29 years old	0.79 (0.39, 1.60)	0.56 (0.22, 1.40)
≥ 30 years old	0.50 (0.22, 1.12)	0.82 (0.29, 2.28)
Education		
< High school	1.15 (0.51, 2.59)	1.51 (0.59, 3.90)
High school	0.80 (0.35, 1.84)	1.46 (0.56, 3.77)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	1.40 (0.65, 3.00)	0.67 (0.26, 1.71)
Hispanic	0.72 (0.31, 1.68)	0.57 (0.21, 1.55)
Other[b]	1.16 (0.38, 3.55)	0.16 (0.02, 1.09)
Number of Living Children		
0	1.00	1.00
1	0.64 (0.33, 1.22)	1.07 (0.52, 2.19)
≥ 2	0.99 (0.45, 2.15)	1.01 (0.45, 2.28)
Marital Status		
Married	1.00	1.00
Not married	1.47 (0.81, 2.70)	1.13 (0.53, 2.41)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	1.29 (0.70, 2.37)	0.73 (0.37, 1.42)
Child		
Birthweight		
< 2,500 grams	0.49 (0.19, 1.30)	1.17 (0.45, 3.06)
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.88 (0.74, 4.77)	0.94 (0.34, 2.64)
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	1.12 (0.65, 1.92)	1.08 (0.58, 2.01)

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.57 (0.31, 1.04)	1.87 (0.93, 3.77)
Receipt of Maternity Care Coordination Services		
Yes	0.80 (0.45, 1.42)	1.01 (0.49, 2.07)
No	1.00	1.00
Receipt of WIC		
Yes	0.72 (0.39, 1.35)	0.77 (0.33, 1.79)
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.07 (0.58, 1.97)	1.17 (0.53, 2.58)
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	0.46 (0.21, 0.99)	1.29 (0.42, 4.01)
Northeastern	0.70 (0.28, 1.75)	0.50 (0.18, 1.35)
Southeastern	0.73 (0.28, 1.94)	1.21 (0.41, 3.53)
Eastern	0.95 (0.37, 2.42)	1.16 (0.36, 3.76)
Western	0.73 (0.28, 1.90)	1.04 (0.40, 2.68)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	0.90 (0.44, 1.82)	0.80 (0.33, 1.94)
Noncore adjacent	1.10 (0.46, 2.60)	1.14 (0.42, 3.05)
Noncore areas not adjacent	1.01 (0.31, 3.37)	0.59 (0.13, 2.67)

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 2.15. Ordinary least squares regression of selected characteristics on home health costs among children with and without orofacial clefts with home health costs during the first year of life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=101)** Cost Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=155)** Cost Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	2.59 (0.79, 8.49)	1.09 (0.65, 1.83)
21-24 years old	1.00	1.00
25-29 years old	1.61 (0.63, 4.08)	0.89 (0.51, 1.53)
≥ 30 years old	0.73 (0.19, 2.78)	0.95 (0.55, 1.64)
Education		
< High school	0.92 (0.28, 3.00)	0.68 (0.39, 1.18)
High school	1.26 (0.44, 3.65)	0.61 (0.36, 1.05)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	2.15 (0.77, 5.97)	1.11 (0.71, 1.72)
Hispanic	0.71 (0.21, 2.47)	0.88 (0.45, 1.75)
Other[b]	4.34 (0.82, 23.10)	0.64 (0.25, 1.64)
Number of Living Children		
0	1.00	1.00
1	4.01 (1.55, 10.36)*	0.70 (0.45, 1.10)
≥ 2	9.99 (2.55, 39.03)*	1.09 (0.63, 1.87)
Marital Status		
Married	1.00	1.00
Not married	0.44 (0.18, 1.05)	1.00 (0.67, 1.51)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	0.51 (0.21, 1.25)	0.96 (0.63, 1.47)
Child		
Birthweight		
< 2,500 grams	5.42 (1.75, 16.80)*	1.98 (1.19, 3.29)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	0.16 (0.04, 0.54)*	1.15 (0.69, 1.91)
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	3.57 (1.54, 9.28)*	0.91 (0.62, 1.34)

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.39 (0.16, 0.92)[*]	0.85 (0.57, 1.25)
Receipt of Maternity Care Coordination Services		
Yes	1.35 (0.59, 3.10)	1.41 (0.91, 2.19)
No	1.00	1.00
Receipt of WIC		
Yes	0.31 (0.13, 0.75)[*]	1.07 (0.67, 1.72)
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.98 (0.59, 3.10)	1.10 (0.71, 1.71)
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	5.59 (1.69, 18.43)[*]	1.11 (0.65, 1.88)
Northeastern	3.31 (0.90, 12.21)	1.88 (0.86, 4.13)
Southeastern	1.24 (0.38, 4.05)	1.06 (0.59, 1.91)
Eastern	1.67 (0.44, 6.31)	0.52 (0.23, 1.16)
Western	1.80 (0.51, 6.41)	1.26 (0.67, 2.36)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	1.28 (0.48, 3.40)	1.12 (0.65, 1.94)
Noncore adjacent	0.50 (0.14, 1.72)	1.14 (0.50, 2.60)
Noncore areas not adjacent	0.32 (0.03, 3.16)	1.63 (0.44, 5.96)

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 2.16. Ordinary least squares regression of selected characteristics on total health costs among children with and without orofacial clefts with total health costs during the first year of life in North Carolina, 1995-2002[a]

Characteristics	Children with Orofacial Clefts (n=563) Cost Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts (n=5,637) Cost Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	0.88 (0.68, 1.14)	1.00 (0.95, 1.04)
21-24 years old	1.00	1.00
25-29 years old	0.90 (0.70, 1.15)	0.98 (0.94, 1.03)
≥ 30 years old	0.75 (0.56, 1.01)	0.98 (0.92, 1.03)
Education		
< High school	1.42 (1.08, 1.88)*	1.10 (1.04, 1.16)*
High school	1.14 (0.88, 1.48)	1.04 (0.99, 1.10)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	0.81 (0.63, 1.05)	0.98 (0.94, 1.02)
Hispanic	0.83 (0.62, 1.12)	0.96 (0.91, 1.02)
Other[b]	1.25 (0.77, 2.03)	1.00 (0.91, 1.10)
Number of Living Children		
0	1.00	1.00
1	0.97 (0.78, 1.22)	0.96 (0.92, 1.01)
≥ 2	0.98 (0.76, 1.27)	0.94 (0.89, 0.99)*
Marital Status		
Married	1.00	1.00
Not married	0.89 (0.73, 1.09)	1.06 (1.02, 1.10)*
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	1.08 (0.87, 1.36)	0.93 (0.89, 0.97)*
Child		
Birthweight		
< 2,500 grams	1.60 (1.14, 2.25)*	2.05 (1.90, 2.21)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.52 (1.08, 2.13)*	1.69 (1.58, 1.82)*
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	1.09 (0.91, 1.31)	1.18 (1.14, 1.22)*

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.90 (0.73, 1.12)	0.98 (0.94, 1.02)
Receipt of Maternity Care Coordination Services		
Yes	1.24 (1.00, 1.54)	1.07 (1.03, 1.11)[*]
No	1.00	1.00
Receipt of WIC		
Yes	0.87 (0.69, 1.08)	1.05 (1.01, 1.10)[*]
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.22 (0.99, 1.51)	1.17 (1.12, 1.22)[*]
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	1.06 (0.80, 1.42)	1.12 (1.06, 1.18)[*]
Northeastern	1.07 (0.79, 1.43)	1.09 (1.03, 1.15)[*]
Southeastern	1.27 (0.92, 1.76)	1.03 (0.98, 1.10)
Eastern	1.00 (0.74, 1.34)	1.03 (0.97, 1.09)
Western	0.93 (0.66, 1.32)	1.10 (1.03, 1.18)[*]
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	1.02 (0.80, 1.29)	1.02 (0.98, 1.07)
Noncore adjacent	0.69 (0.49, 0.97)[*]	1.03 (0.96, 1.09)
Noncore areas not adjacent	1.17 (0.70, 1.95)	1.08 (0.99, 1.18)

[a] All children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Figure 2.1.

Percentage of Total Service Use by Service Category for Children with Orofacial Clefts During the First Year of Life in North Carolina, 1995-2002

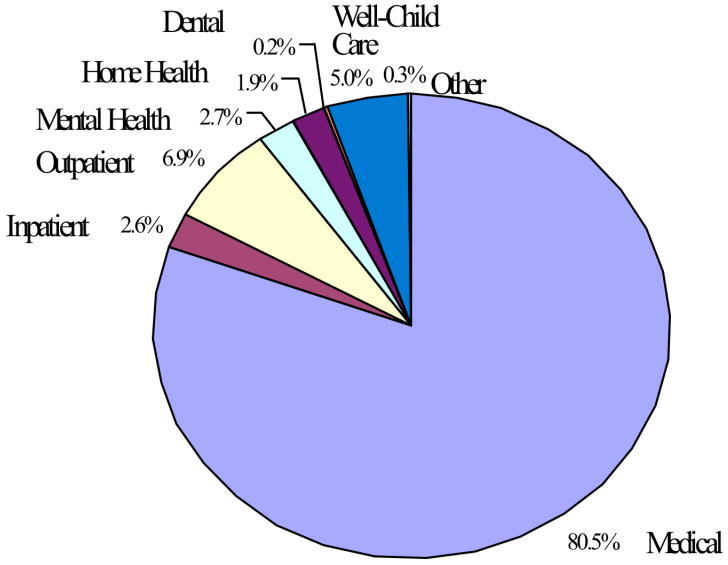


Figure 2.2

Percentage of Total Services by Service Category for Children without Orofacial Clefts in North Carolina, 1995-2002

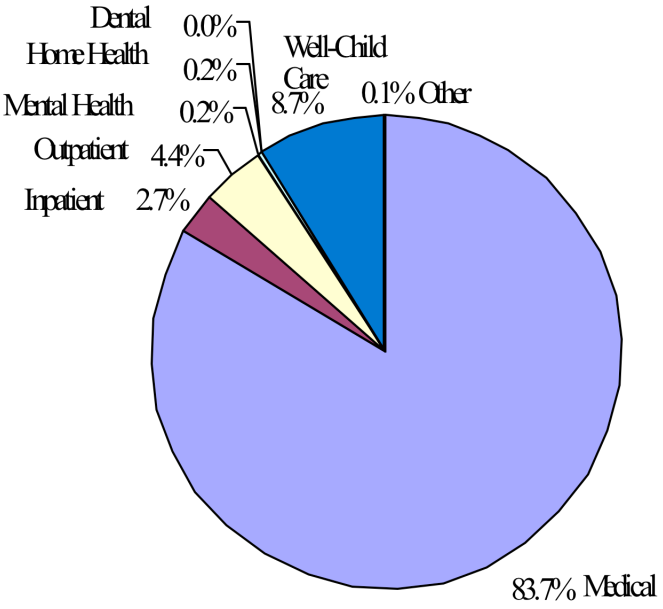


Figure 2.3

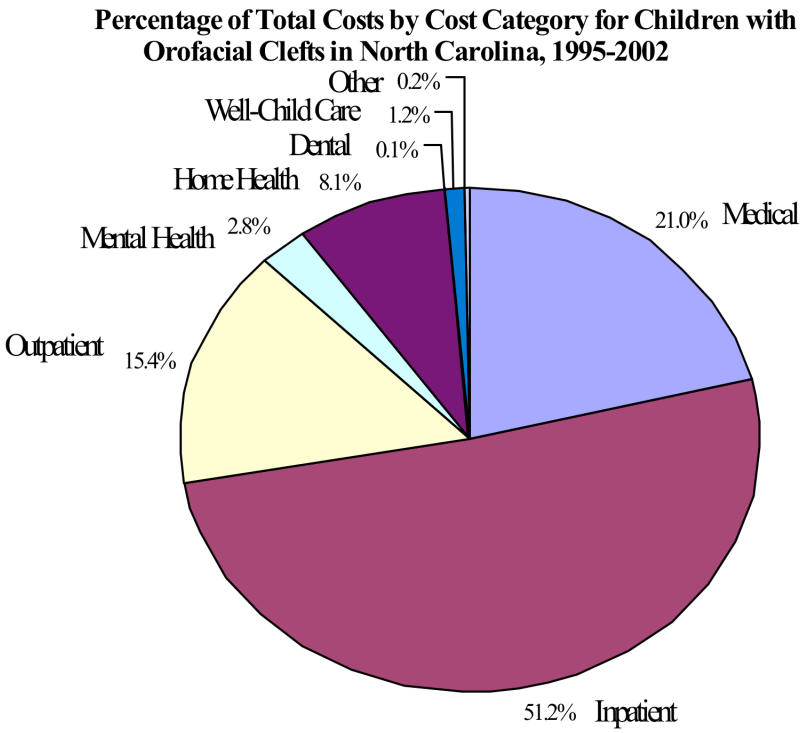
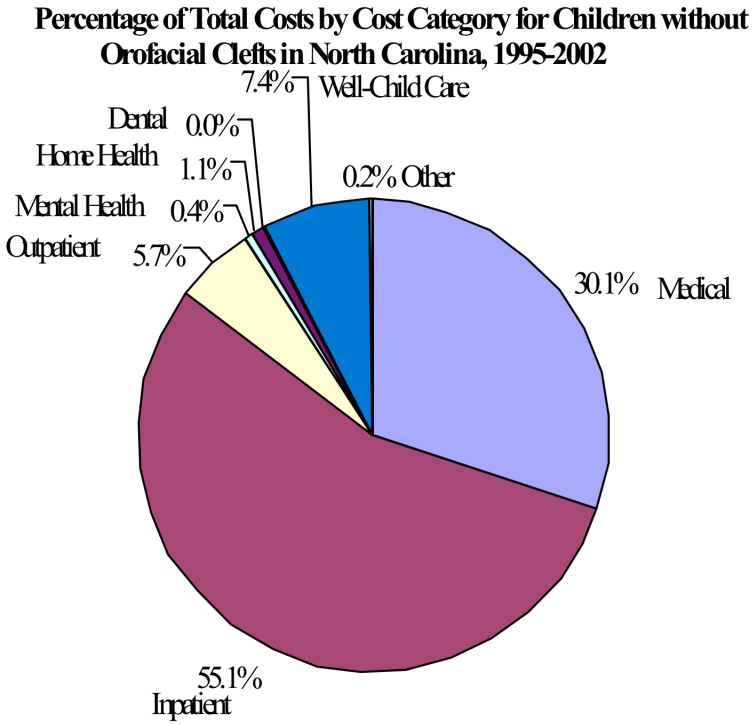


Figure 2.4



CHAPTER III

TIMELINESS OF SERVICES DURING THE FIRST TWO YEARS OF LIFE AMONG CHILDREN WITH OROFACIAL CLEFTS IN NORTH CAROLINA, 1995-2002

Abstract

Background and Objectives: In 1993, the ACPA developed parameters of care for patients with craniofacial conditions such as OFC. To date, no study has examined the timeliness of services according to these guidelines. The objectives of this study were to determine: 1) proportion of children with OFC who received primary cleft surgery within the first 18 months of life; 2) factors associated with timely cleft surgery; and 3) proportion of children who received selected specialized services during the first two years of life among children with OFC per the ACPA guidelines.

Methods: Data from the North Carolina vital statistics, birth defects registry, and Medicaid enrollment and paid claims were linked to identify resident children with OFC born 1995-2002 who were continuously enrolled in Medicaid through age two (N=406). Proportions of children who received primary cleft surgery within 18 months of life and age in months when surgery occurred by cleft type and presence of other birth defects were determined. Proportions of children who received specialized service such as speech and language therapy within ACPA guidelines were also determined. Multivariate logistic regression analysis was used to examine associations between selected maternal, child, and system characteristics and timely cleft surgery.

Results: About 78% of children with OFC had primary cleft surgical repair within 18 months of life. Of those children who received timely surgery, the mean age surgery occurred was five months. About 51% of children received speech and language therapy, 28.3% received audiology services, and 24.4% received otolaryngologic care within the ACPA recommendations. After adjusting for selected characteristics, children whose mothers received MCC, received PNC at a local health department, or lived in the southeastern or northeastern region of the state were strongly associated with receipt of primary cleft surgery within 18 months of life.

Conclusion: Most children with OFC who were continuously enrolled in Medicaid received primary cleft surgery within the ACPA recommendations. Many children did not receive other necessary specialized services. Efforts to increase timely receipt of services for this population to improve their health outcomes are needed.

Background

Orofacial clefts can impair the development of teeth, speech, hearing, feeding capabilities, and psychomotor and cognitive skills, thereby creating physical and emotional stress for infants, children, and their families. Timing of medical and ancillary services and treatment are imperative for good health outcomes, particularly with feeding, speech, hearing and dental/orthodontic outcomes among children with OFC (6-15, 109, 191, 192). Medical services refer to the treatment of OFC such as closure of the cleft and secondary conditions and ancillary services refer to allied health and related professional services usually provided in rehabilitation, education, community-based clinic settings, or home health.

Services and treatment for children with OFC can vary depending on cleft severity, presence of associated syndromes and/or another birth defect, and the child's age and needs (8, 109). For example, a child who has CL only may not need as many services compared to a child with CLP, associated syndrome, and another birth defect (8).

Despite these variations, some general recommendations exist for services and treatment for this population (8, 109-111). These recommendations were originally set forth by the ACPA in 1993 and were amended in 2000 and late 2004 (109). Initial evaluations of infants with OFC are recommended within the first few days of life and subsequent evaluations should be scheduled at regular intervals. The frequency of evaluations is contingent on the cleft severity and child's age.

At the first visit, a full pediatric evaluation, including nutritional and feeding assessments and a medical history, should be conducted. Genetic / dysmorphology screening and subsequent referrals for complete genetic evaluations should also be conducted at the initial visit. Infants and children with OFC need genetic services to determine the presence of associated syndromes, complexity of care involved, and the family and child's needs such as further evaluations and/or recommended treatment (8). Undetected associated syndromes or other birth defects can increase the risk of developmental disabilities, speech disorders, and airway comprise (8). Other evaluations that should occur during this time include surgical, otolaryngologic, audiologic, prelinguistic speech-language, psychosocial, and dental. Primary cleft surgical repair of the lip is necessary to reconstruct the normal anatomy and function of the lip, correct the nasal deformity, and construct the floor of the nose and proper alignment of the gum-line. Primary cleft surgical repair of the palate or palatoplasty is to close the palatal defect and create an adequately functioning velopharyngeal mechanism for normal

speech production. The outcome of palatoplasty is critical to speech, facial growth and eustachian tube function. Both surgical cleft repairs can help improve speech, language, dental, and psychosocial outcomes. For example, children who have their palate closed before the age of one, usually develop normal speech earlier and easier than children who have their palate closed after that age (9).

Otolaryngologic and audiologic services are recommended because of increased hearing loss during infancy for children with OFC (8). Dental services are recommended due to the increased risk of dental caries (11, 112-114). Well-child care or preventive services are also recommended to help establish a community medical home that provides continuity of care that is accessible, culturally competent, and compassionate as promoted by the Academy of Pediatrics and the National Center of Medical Home Initiatives for CSHCN (8, 116, 117, 119, 121, 123, 193).

Families of children with OFC should also receive advice and information on special feeding devices, speech difficulties of children with compromised airways, and speech therapy and language impairments. In addition, families should be given information on otologic disease symptoms, hearing loss, sleep apnea, oral cavity care, caries prevention, and contacts for appropriate support groups and case coordinators.

Per the 2000 ACPA guidelines, speech and language, audiological, and dental services are recommended within the first year of life. Surgical closure of the CL should occur within the first six months of life and closure of the palate should occur within 18 months of life (109). If a child is diagnosed with both CLP, primary surgical repair should occur within 18 months of life. Otolaryngological services are recommended within the first six months of life. Genetic services should occur within the first two years of life, and psychological services

are recommended periodically through adolescence (109). These recommendations slightly changed in 2004. Audiological services are recommended within the first three months of life, and primary CL surgical repair is recommended within the first 12 months of life. Table 3.1 lists an abbreviated version of the 2000 ACPA recommended services and treatment and frequency of these services for children with OFC (8, 109).

As demonstrated, the ACPA recommends services and treatment throughout childhood and adulthood for children with OFC. Receipt of these services, including timeliness, is essential to the medical and psychosocial well-being of children with OFC. Improving health outcomes of children with OFC is one of CDC's priority research areas for this population (33). Despite recommended services and treatments, no study has assessed the timeliness of services in accordance with nationally recommended treatment guidelines for children with OFC.

The primary objective of this study was to determine the percentage of children with OFC who received primary cleft surgical repair within the first 18 months of life and the mean age at which cleft surgery occurred. These results were stratified by cleft type and presence of other birth defects. Another objective was to examine the effect of selected maternal, child, and system characteristics on the receipt of timely primary cleft surgery. The last objective was to examine the percentage of children who received other specialized services within the ACPA recommendations by cleft type. It was hypothesized that the majority of children with isolated CP and children with CLP would have their primary surgical repair within 18 months of life, and the majority of children with CL would have their primary surgery within the first six months of life. It was also hypothesized that children with CL alone or children with CLP would be more likely to receive timely cleft surgery than children with isolated

CL. It was also suspected that families of children traveling less than or equal to 30 minutes to a craniofacial center or team would be more likely to receive timely cleft surgery than families of children traveling greater than or equal to 90 minutes. Lastly, it was hypothesized that the majority of children with OFC would receive specialized services within the ACPA recommendations.

Methods

Study Design and Sample

This study was a retrospective, cohort study of North Carolina resident children with OFC born between 1995 and 2002 and who were continuously enrolled in Medicaid during the first two years of life. Children with OFC were ascertained by the NCBDMP using British Pediatric Association codes 749.000-749.290 (n=1,355). Infants who died within the first 12 months of life (n=103), who were born out of state, or who were adopted were excluded from the study.

Using the birth certificate number, children with OFC identified from the NCBDMP were linked with the BabyLove files to identify which children were on Medicaid. The matching rate was 100%. Using the infant Medicaid identification number, children were matched from the BabyLove files to the Medicaid enrollment records to identify children who were continuously enrolled in Medicaid during the first year of life (n=565) and during the first two years of life (n=406). The matching rate was 98.5%. Continuous enrollment in Medicaid was defined as enrollment greater than or equal to 11 months per year of life. This is a commonly used definition for continuous enrollment in Medicaid and utilized as the standard definition for the Health Plan Employer Data and Information Set, which is the most widely

used set of performance measures in the managed care industry (66, 144, 145). Enrollment records were then matched to Medicaid paid claims to determine timeliness of services, which yielded a matching rate of 100.0%. Records of all services received and paid for by Medicaid for calendar years 1995-2004 were extracted to allow for two years of health service utilization data for all children with OFC born during the study period.

Data Sources Utilized and Variable Construction

Data sources for this study included the North Carolina BabyLove files, NCBDMP, and Medicaid enrollment records and paid claims. The BabyLove files are composite North Carolina birth files that contain information on vital statistics, child service coordination, WIC, Medicaid status, PNC source, and MCC services. The NCBDMP is a population-based, statewide program that includes all resident live births, fetal deaths, and therapeutic abortions at any gestational age. The program covers all of North Carolina hospitals and approximately 120,000 resident births per year. Most major birth defects are ascertained, including over 200 types of structural defects, and infants are ascertained up to one year after delivery.

The primary outcomes of interest were timely receipt of primary cleft surgery, otolaryngologic care, audiological services, speech and language therapy, dental services, psychological and social services, and genetic services. To determine timeliness of services, service receipt dates were employed. In this study, timeliness was defined using the 2000 ACPA recommendations through age two as these guidelines were most applicable to the study population. For children with OFC, timely primary cleft surgery was defined as surgery within 18 months of life. By cleft type, timely CL surgery was defined as surgery within the first six months of life and timely CP with or without CL was defined as surgery within 18

months of life. Timely receipt of otolaryngologic care was within the first six months of life and once per year through age two. Timely audiological assessments, speech and language therapy, dental services, and psychological and social services, including screening evaluations were defined as within the first year of life and once per year through age two. Timely genetic screening, including follow-up evaluations, were defined as once within the first two years of life (109).

To create these outcomes using the Medicaid paid claims, all past and current codes from the *Physicians' Current Procedural Terminology* and diagnostic-related group codes for these services were employed. For dental services, the Medicaid paid claim pre-established category for dental claims and all past and current procedural terminology and diagnostic-related group codes related to dental and orthodontic care were used. To ensure a comprehensive inclusion of procedural codes used during the study period, consultations occurred with several members of craniofacial centers and teams in the state. Any procedural codes for these services used by the craniofacial centers and teams and the Children's Developmental Services Agency for reimbursement of Medicaid in the state were also included. Children's Developmental Services Agency implements the Infant-Toddler Program in each region of North Carolina, which is the state's interagency system of early intervention services for children aged birth to five years old with special health care needs. Certain procedural codes were excluded because these were standard codes used for generic office visits and the specific service rendered could not be determined. These codes were 99202, 99212, 99213, and 99214. However, if from the provider specialty code the specific service rendered could be determined, these codes were utilized.

Characteristics possibly associated with the receipt of timely cleft surgery were categorized as maternal, child, and system characteristics. Maternal characteristics included age, number of living children, race/ethnicity, education, marital status, and initiation of PNC in the first trimester. Child characteristics included birth weight, preterm birth, gender, cleft type, and presence of other anomalies. System characteristics consisted of source of PNC, receipt of MCC services, receipt of WIC, birth hospital level of care, perinatal care region, place of residence, and travel time to closest craniofacial center or team.

Two geographical variables, perinatal care region and place of residence, were analyzed to determine any geographical differences in receipt of timely services. Perinatal care regions are based on population and county locations. They were geographically defined as northwestern, northeastern, southwestern, southeastern, western, and eastern. These regions were established in the 1980's in an effort to develop regional referral networks of perinatal care in North Carolina (148, 149). Urban Influence Codes were utilized to classify counties into 12 levels that build on the Office of Management and Budget metropolitan and nonmetropolitan dichotomy (150-154). In this study, the 12 levels were collapsed into four categories based on metropolitan, nonmetropolitan, adjacency, and non-adjacency areas.

Adjacency included physical adjacency and at least 2% of the population commuting to the metropolitan or micropolitan area (154).

To determine time traveled to the closest craniofacial center or team, the maternal birth address was used from the BabyLove files in the GIS analysis. For the GIS analysis, geocoding was performed all on children with OFC who were continuously enrolled in Medicaid through age two. The GIS analysis was conducted using *ESRI ArcGIS* and *Network Analyst 9.2. TeleAtlas Multinet 2005 2.1* was used for the road network to match an address

on a street network to obtain real-world coordinates and to be able to place that address on a map. The street network is attributed with address ranges, street names, street types, cities, and zip codes. The software then performed a "match" against the road reference and interpolated where along a road to place the point. Spatial analysis of point-to-point distance was also performed within *ArcGIS*, which provided the average distance and time traveled between residential addresses of families of children with OFC and the two craniofacial centers and five craniofacial teams in North Carolina. Craniofacial centers are located in Winston-Salem and Chapel Hill, and craniofacial teams are located in Charlotte, Winston-Salem, Durham, and Greenville (Appendix H).

Statistical Analyses

All analyses were conducted on children with OFC who were continuously enrolled during the first year of life or during the first two years of life. Relationships between maternal, child, and system characteristics and cleft type were assessed using Pearson chi-square if cell sizes were greater than five or Fisher's exact tests if cell sizes were less than five.

Bivariate and stratified analyses were performed on the receipt of timely primary cleft surgery and maternal, child, and system characteristics. To determine the magnitude of these associations and precision of these estimates, crude odds ratios (OR) and 95 percent confidence intervals (CI) for the odds of receiving timely cleft surgery by maternal, child, and system characteristics were computed. Multivariate logistic regression analysis was employed to determine the effect of selected maternal, child, and system characteristics on the receipt of timely primary cleft surgery. To determine the magnitude of these associations and precision of these estimates, crude and adjusted OR and 95 percent CI were computed.

To determine the most parsimonious model, backward elimination modeling and a p-value of 0.10 for the likelihood ratios tests were employed to determine which maternal, child, and/or system factors contributed significantly to the timely receipt of primary cleft surgery. In the bivariate and multivariate analyses, timely cleft surgery for all OFC was categorized as a binary outcome, less than 18 months life or greater than or equal to 18 months of life.

Using Pearson chi-square or Fisher's exact tests with a p-value less than 0.05 for statistical significance or Breslow- Day test of homogeneity with a cut point of p-value less than 0.10, effect modification by cleft type was assessed. To determine if cleft type modified the relationship between presence of other anomalies and timely cleft surgery, stratum-specific odds ratios were examined and a cut-point of p less than 0.10 was used in the Breslow-Day test of homogeneity. Effect modification also was assessed in the associations between maternal, child, and system characteristics and timely receipt of otolaryngologic care, audiological assessment, speech and language therapy, dental services, psychological and social services, and genetic services.

From the 565 children with OFC who were continuously enrolled in Medicaid during the first year of life, six children had missing data for one or more of the variables analyzed. Because this represented 1.1% of the entire dataset, missing data had very little effect on the results. Consequently, these children were not deleted from the data set; however, they were deleted in the analyses. All analyses were conducted using SAS software, version 9.1.

This study was approved by the University of North Carolina at Chapel Hill Public Health Institutional Review Board, North Carolina Division of Public Health Institutional Review Board, and the North Carolina Division of Medical Assistance.

Results

Sample Population Characteristics

For children with OFC continuously enrolled in Medicaid during the first year of life and through age two, the distributions of all maternal, child, and system characteristics among the cleft types were similar except maternal education, child's gender, and presence of other anomalies. Children with CLP were significantly more common among mothers with less than a high education and male children. Children with isolated CP were more common among mothers with a high school education. About 65% of the study population had isolated clefts, and multiple anomalies were significantly more common among children with CP. About 15% more children with OFC were born in a community hospital than in a tertiary care center, but this difference was not statistically significant. The percentages of mothers receiving MCC services, WIC, and PNC at a health department were higher among study participants than all mothers giving birth in North Carolina during this time period (Table 3.2). The only characteristics that differed among the cleft types for children who were continuously enrolled in Medicaid during the first year of life and through age two were number of living children and maternal age.

Of the 565 children with OFC continuously enrolled in Medicaid during the first year of life, 80.4% (n=454) of children were geocoded, and of the 406 children with OFC continuously enrolled in Medicaid through age two, 77.8% (n=316) of children were geocoded. About 20% of children in both groups could not be geocoded due to post office box addresses or rural routes.

Timeliness of Primary Cleft Surgery and Specialized Services

About 78% of children with OFC received primary cleft surgical repair within 18 months of life. Among children with OFC who received timely primary cleft surgery, the mean age at which surgery occurred was five months. About 88% of children with CL received primary surgery within the first six months of life and 58.0% of children with CP received primary surgery within 18 months of life. The average age primary cleft surgery occurred was similar for children with CL with or without CP, about three months. Children with isolated clefts were 15% more likely to receive surgery within 18 months of life than children with multiple anomalies (Table 3.3).

During the first year of life, speech and language therapy was the most commonly used service (51.1%) followed by audiology services (28.3%) and otolaryngologic care (24.4%). Children with CL had the lowest percentage of receipt of these specialized services (Table 3.4). Very few children with OFC received timely otolaryngologic care, audiology services, speech and language therapy, dental, genetic, and psychological services according to the ACPA recommendations (Table 3.5). About 51% of children received speech and language therapy within the first year of life and 33.0% received this service in both the first and second year of life. Only 7.8% of children with OFC received otolaryngologic care within the first six months of life, and only 10.8% of children received this type of care in both the first and second year of life. Only 5.2% of children with OFC received dental care in both the first and second year of life (Table 3.5).

There was heterogeneity across cleft type for timely receipt of speech and language therapy and birth hospital level of care (p-value=0.06) and perinatal care region (p-value=0.05). (Data not shown.) A child's cleft type did not modify the relationship between

timely receipt of audiological or otolaryngologic services and maternal, child, and system characteristics. Data were generally sparse, precluding further investigation of effect modification across cleft type for timely receipt of dental, genetic, and psychological services and maternal, child, and system characteristics.

Predictors of Timely Primary Cleft Surgery

Because not all children with OFC could be geocoded, multivariate logistic regression models were conducted among the 316 children with OFC who were continuously enrolled in Medicaid through age two and who were geocoded. Separate models were conducted that included and excluded the time traveled variable to determine if it contributed significantly to the model. Using the likelihood ratio test, it was found that the time traveled variable did not contribute significantly to the model (p-value=0.16). In addition, the effect estimates for the other characteristics remained stable. Consequently, this variable was not included in further multivariate analyses and was not included in the crude and adjusted OR results in Table 3.6. Because cleft type did not modify the relationship between presence of other birth defects and timely primary cleft surgery, an interaction term of cleft type and presence of other anomalies was not included in the multivariate regression models.

After adjusting for maternal, child, and system characteristics, mothers who received MCC or PNC at a health department or lived in the southeastern or northeastern region of the state had strong positive associations with their children receiving primary cleft surgery within 18 months of life (Table 3.6). Children with OFC whose mothers received MCC were about twice as likely to have their children receive timely primary cleft surgery compared to those whose mothers did not receive such services (adjusted OR: 2.38; CI: 1.16, 4.89).

Multivariate logistic regression results also indicated racial/ethnic and geographical differences in the receipt of timely cleft surgery among children with OFC. Children of Black/non-Hispanic mothers were 70% less likely and children of Hispanic mothers were 14% less likely than children of White/non-Hispanic mothers to receive surgery within 18 months of life. Children living in the northeastern and southeastern region were 153% and 54% respectively more likely to receive timely cleft surgery than children living in the northwestern region of the state. In contrast, children living in the southwestern region were 66% less likely to receive surgery within 18 months of life. Children living in noncore areas adjacent to a small metropolitan or micropolitan area were 77% less likely to receive timely cleft surgery than children living in a metropolitan area (Table 3.6).

The most parsimonious model included maternal age and race, cleft type, presence of other anomalies, child's gender, receipt of MCC, perinatal care region, and place of residence. Due to small sample sizes for cleft type, factors associated with timely primary cleft surgery by cleft type could not be determined.

Discussion

The results from this study show that most children with OFC continuously enrolled in Medicaid in North Carolina who were born between 1995 and 2002 received primary cleft surgical repair within the ACPA recommendations. These results substantiated our hypothesis. In contrast, most children did not receive other necessary, recommended specialized services during the first two years of life, which was counter to our hypothesis.

Compared to all live births in North Carolina born during the study period, children with OFC had a higher proportion of children born LBW and preterm. This was to be expected as

children with OFC are at an increased risk of being born LBW and/or preterm, especially if they have an associated syndrome and/or another birth defect (160, 161). This study also found that the majority of children had isolated clefts and children with multiple anomalies was more common among children with CP, which was to be expected (3, 161). As found in previous studies, this study found a higher proportion of isolated CP in female children and an increased percentage of CL with or without CP among male children (35, 46, 47, 129). Unlike in previous studies, this study had a higher proportion of children in each cleft group among White/non-Hispanic than Native Americans and Asians. However, this may be due to more White/non-Hispanic mothers than Native Americans and Asians on Medicaid during the study period. The percentages of mothers receiving MCC services, WIC, and PNC at a local health department were higher among study participants. This result was to be expected among mothers receiving Medicaid because mothers can only receive MCC if they are Medicaid-eligible. These results may also be due to having a referral system in place for mothers receiving MCC.

Contrary to our hypothesis, when travel time was included in the multivariate model, children whose families traveled greater than or equal to 90 minutes were 4.7 times more likely to receive timely primary cleft surgery than children whose families traveled less than or equal to 30 minutes (adjusted OR: 5.70 95% CI: 1.08, 30.07). (Data not shown.) This effect estimate was most likely unreliable due to wide confidence intervals, so it must be interpreted with caution. Regardless, this finding was congruent to a recent study by Rodd et al. on failed attendances at pediatric dental, multidisciplinary cleft clinics, and orthodontic clinics among children with and without OFC. The authors found that distance traveled to the clinics had no significant effect on attendance rates (194).

Results from this study indicated racial/ethnic and geographical differences in the receipt of timely primary cleft surgery and specialized services among children with OFC. This may have been due to the availability or unavailability and knowledge of craniofacial centers and teams in the state. Racial and ethnic disparities in service utilization and lack of access to care among children are well-known, especially for Black/non-Hispanic and Hispanics (145, 195-198). These minority groups may have been unaware of the specialized services needed to treat their child and/or unaware of the craniofacial centers and teams in the state. Surprisingly, the southwestern perinatal care region was the least likely to have children receive timely primary cleft surgery; yet, this region has two craniofacial teams.

Geographical differences in timely receipt of specialized services, including primary cleft surgery, for children with OFC may be due to location of the craniofacial center and team and the service types available at a craniofacial center or team. In North Carolina, craniofacial teams and centers are available in certain geographic locations and thus lacking in other geographical locations (199). Despite having seven craniofacial centers and teams in North Carolina, covering more geographical areas than states in the mid-west, there are none in the southeastern and western perinatal care regions of the state (Appendix H). This affects over 13% (n=140) of families of children with OFC on Medicaid and not on Medicaid born during 1995-2002. This means these families could be receiving coordinated services from a team of health care providers specializing in craniofacial care closer to home.

Differences in services and treatment between and within craniofacial centers and teams exist, which can affect timeliness of services (126). For example, some craniofacial teams may provide no direct clinical treatment, but only evaluation and quality assurance. In comparison, craniofacial centers usually provide more services and treatment. Differences

may also exist in the number and types of services the centers and teams offer such as some centers and teams offer dental and orthodontic care whereas some may not. Craniofacial centers and teams vary in their capacity to treat patients with clefts and/or other craniofacial conditions as well (126, 199). Another difference is that some centers or teams may not accept Medicaid patients or new patients. Despite the ACPA recommendations, differences can exist between the plastic and oral and maxillofacial surgeons regarding treatment options with pre and post-surgical orthodontic appliances and with presurgical orthopedics like with nasal moldings in infancy. Nasal moldings adjust the cleft position segments into a more ideal environment prior to primary CL repair surgery and can make lip repair easier for the plastic surgeons (200). All these factors could have affected the timeliness of services among children with OFC.

Our results may also be due to parental perception of need and cleft severity. Orofacial clefts, especially CL, are readily apparent at birth. This type of birth defect compared to other more severe birth defects can be readily and sometimes easily treated, especially CL alone. Consequently, parents usually see and understand the need to medically repair the cleft. Parents may place a higher priority on the more surgical, medical needs such as repairing the cleft rather than obtaining ancillary services like dental, speech, language, and audiology that children might need (98, 133-136).

Low dental service use may be due to parents not recognizing the need for dental care for their children with OFC (165). Additionally, most children do not see a dentist during infancy (112, 163). However, for children with OFC it is important to see a dentist in the first year of life due to the need of specialized oral health services and higher risk of a variety of oral health conditions such as dental caries and periodontal disease (11, 112-114). This is a

need that is clearly not being met and was recently recognized as such by the American Academy of Pediatric Dentistry (130). Low dental use could also be explained by low or no reimbursement from Medicaid for specialized dental and orthodontic care of children with OFC (126, 130-132).

Some parents may be unaware of the need for medical and ancillary services for their child, including the need for cleft repair surgery, which can delay care and can result in poor health outcomes (201, 202). Previous studies have shown that lower income and less educated parents are less likely than higher income and more educated parents to say their CSHCN needed specialized services to treat their children (201). Parental perception of the lack of need for specialized services among children with OFC may be particularly true among children with bivid uvula and/or submucous CP, which are minor forms of CP. Children with these minor clefts may not have had primary cleft repair surgery during the first two years of life, if at all (203, 204). This is because these minor clefts may not have been diagnosed until later in life, if at all, and some children with these minor clefts may never need surgery. Unfortunately, this study could not determine the percentage of children OFC who had such minor clefts. However, these minor clefts are rare in the general population, for example, the prevalence of bivid uvula is 0.13 per 1,000 children and submucous CP occurs one in 10,000-20,000 children (9, 204).

Even though psychological and social services are recommended by the ACPA within the first year of life, most children do not need and receive them until early childhood to assist with psychosocial, social interaction, and self-image problems (8). In addition, like with dental care, parents may not think their child needs psychological and social services during the first year of life. As such, low use of psychological services was expected in this study.

Other possible reasons for our results include parents already being familiar with the health care system and/or in a referral system during the prenatal and postnatal period. A cornerstone of MCC services are to develop a strong referral network, assist in accessing resources and ensure appropriate services are rendered, including continuity of care (162). It also may be explained by most of the craniofacial centers and teams in North Carolina are affiliated with a tertiary care centers (6, 124, 125). Children being born at a tertiary care center could be directly referred to a craniofacial center or team at that hospital. This would contribute to parents' knowledge of the health care system and/or knowledge of craniofacial teams and centers that specialize in cleft surgery and delivery of craniofacial care. Previous studies found that identification of children with OFC and referral to services, especially to craniofacial centers and teams were significantly associated with areas of residence, cleft type, presence of other birth defects, presence of other malformations in the family, and receipt of MCC services (32, 129, 205).

However, despite the *Healthy People 2010* objectives 21-15 and 21-16 of promoting efforts to increase the number of states that have a referral system in place to refer children with OFC to craniofacial rehabilitative teams and to conduct craniofacial health surveillance, only 23 states and the District of Columbia had such systems in place in 1997 (139). A recent North Carolina study substantiated this fact and that many children with OFC were not referred to child service coordination programs. The authors found that only 45% of children with OFC born 1999-2002 in North Carolina were referred to the Child Service Coordination Program, which is a state-based program that identifies high-risk women and children for services (205). Referral systems can affect receipt and timeliness of services among children

with OFC (32, 129, 205). However, this study did not examine the effect of referrals on service utilization for children with OFC, which was a potential limitation.

Quantitative and qualitative evidence exists on the lack of health care providers trained and experienced in care of children with OFC children at community hospitals and in specialized services such as dental and orthodontic care (8, 164, 166, 206, 207). Some hospitals are better prepared than other hospitals to care for children with OFC due to either being affiliated with a craniofacial team and/or having treated large volumes of patients with this condition (208). As a result, some hospitals and health care professionals may be more aware of or more familiar with specialized bottles and nipples and feeding devices for these children (208). In a recent survey assessing community orthodontists previous training and experience with caring for children with craniofacial disorders such as OFC in Washington state, 80% of respondents had seen less than three patients with CL with or without CP in the past three years (166). In a national survey of United States and Canadian dental schools, about half of the schools in the United States provided students with more than five hours of classroom instruction and less than five percent of clinical time was dedicated to providing care of CSHCN (209). In North Carolina, there is a shortage of Medicaid dental providers, especially the specialty needed to treat children with OFC (164, 166-168). All these factors could contribute to low utilization of specialized services among children with OFC.

It is important to note that our results may be due to how specialized services are being billed by Medicaid or another payor and not necessarily that these children are not receiving these specialized services. Children with OFC on Medicaid may have received services and treatment, but had another payor source and/or went to hospitals where services and treatment were free. At St. Jude Children's Research Hospital in Memphis, Tennessee, and

Children's Hospital of the King's Daughters in Norfolk, Virginia, children with OFC can receive services and treatment for free (202). State-financed and private sources of care such as Shriners Hospitals for Children and Children Healthcare Options Improved through Collaborative Efforts and Services also exist that can pay for services and treatment for children with OFC (210). To the extent this occurred and its affect on these results is unknown.

Strengths and Potential Limitations

There are other factors that can impede receipt of timely services among this population such as financial and non-financial barriers. This study did not examine such barriers, but they are worth mentioning due to their likely impact on the results. Financial barriers among CSHCN are well-known. With Medicaid and other forms of public and private insurance, these barriers could include low reimbursement rates for services, restriction on number of services eligible for benefits such as with speech and language therapy, and denied services and treatment such as with orthognathic and other orthodontic surgeries. Non-financial barriers include structural (structure of the health care system) and personal (situational and psychosocial) barriers (98, 133-136). Structural / system barriers include health policies, service delivery characteristics, delivery system structure, service organization, availability of referral systems to identify and refer children with OFC to craniofacial centers and other specialized services, providers and multidisciplinary teams to treat children with OFC (85, 98, 133-137). Such barriers result from children with OFC, being misidentified, unidentified, or identified later in the developmental stages. Improper identification can be due to cost, the wrong tools being employed to identify high-risk populations, or lack of coordinated care and

can ultimately delay referrals and necessary care (138). Parental lack of knowledge these coordinated care systems and teams exist for children with OFC can also be a structural barrier and can impact timely receipt of care.

Personal barriers can entail parents of children with OFC not knowing Medicaid will pay for certain services or the inability to navigate the health care system and use health care services effectively (98, 133-136, 182). Transportation, timing of appointments, missed work and school days, location of services, no social support system, parental and child attitudes and beliefs such as distrust among health care professionals, knowledge, family dynamics, and cultural factors such as language and cultural norms are also part of personal barriers (98, 133-136).

Results from this study may be due to potential effect modification with cleft laterality and potential confounding factors such as prior use of craniofacial services. Children with bilateral clefts and/or associated syndromes and/or presence and severity of other anomalies will have more complicated health issues than children with isolated clefts, which could affect referral to services and service use. Occurrence of the same birth defect in a family and prior use of certain services could potentially increase access to services and service use, thereby affecting the proportion of children with OFC receiving timely services. Unfortunately, data were unavailable to analyze the effect of cleft laterality and associated syndromes.

During well-child care visits, hearing screening and dental services are supposed to be rendered along with vision and medical check-ups and referrals for treatment. Hence, hearing screening and dental services may be captured under the Medicaid well-child care paid claims. Yet, such services could not be discerned from these paid claims and thus were not

included in the analysis (8, 169, 171, 175, 211). Consequently, this may have led to an underestimation of children with OFC receiving timely dental and otolaryngologic care. With the results from Chapter Two of underutilization of well-child, this was unlikely to affect our results.

Some other potential limitations are exclusion of generic procedural codes for office visits and inadvertently omitting other related procedural codes. Some craniofacial centers and teams in North Carolina utilized generic current procedural terminology codes for specific services such as otolaryngologic or genetic services and pediatric and general dentistry. These codes were excluded from the analysis because the specific services rendered at these visits were unable to be determined. In addition, it is possible that other current procedural terminology and diagnostic-related group codes were inadvertently omitted from the analysis. Some children could have received specialized services, but were not captured in these results, which may have biased the results.

Two geographical variables, perinatal care region and place of residence, were included in this study because each has their own strengths and limitations. Perinatal care regions are more heterogeneous because they consist of urban and rural areas. Yet, they are useful in organizing Title X services and are still used today to organize service delivery within the state. In addition, the regions are a useful way to look at maternal and child health indicators because they are population-based (148, 149). Unlike perinatal care regions, Urban Influence Codes take into account adjacency to urban areas and commuting patterns of populations and are less heterogeneous. Like perinatal care regions, these codes are based on counties, but Urban Influence codes are classified into 12 levels that build on the Office of Management and Budget metropolitan and nonmetropolitan dichotomy (150-154). Urban Influence Codes

are beneficial when examining the structure of health care systems and whether outcomes or care are possibly related to the complexity of the medical or health community (150, 151). This typology is also helpful in distinguishing the effects of adjacency compared to size of the area on access and health service utilization (152). Using Urban Influence Codes with place of residence and perinatal care regions can assist in targeting resources and services to populations in need of services such as in the southwestern and western perinatal care regions and in micropolitan and noncore areas adjacent metropolitan areas or small towns.

With the GIS analysis, about 20% of children could not be geocoded due to rural routes and post office box addresses. This was to be expected due to restricting this analysis to low-income families, which tend to live in more rural areas. Even though selection bias was detected among the children who were geocoded, time traveled to the nearest craniofacial center or team was not a significant predictor of timely cleft surgery; hence, this was not problematic in this study.

Another potential limitation with the GIS analysis is the extent to which families and children actually received services and treatment at the craniofacial centers or teams in the state is unknown. Children may receive services at multiple craniofacial centers or teams. Furthermore, families may not take their child to the closest craniofacial center or team, but to other craniofacial centers and teams in the state where their health insurance is accepted or where the health care providers treat high volumes of children with OFC. In addition, the craniofacial team in Greenville, in the eastern region, was formed during the study period, which would have resulted in families traveling much further to receive care at a craniofacial center or team. Thus, the time traveled results may underestimate the true distance and time traveled to receive craniofacial care. However, this study is the first to examine time traveled

after adjusting for variables such as maternal age and race, birth hospital, and geographic variables such as perinatal care region and place of residence. The last study to examine travel distance to services among this population was in the 1960's and did not adjust for possible confounding variables (32). Also, it was conducted prior to the use of GIS.

This study represents children with OFC on Medicaid, a subpopulation of children in the state, and thus may not be representative of all children with OFC receiving services and treatment. Receipt and timeliness of services can vary by type of health insurance due to different referral policies and reimbursement rates (81, 178). However, using Medicaid data was the only way to examine receipt of specialized services for this population in the state. In addition, it was a strength in that all children in this study were in families of relatively low-income, which provided a more homogeneous population and reduced the potential for confounding by socioeconomic status. In addition, Medicaid paid claims are a bill paying system that require providers to report data for reimbursement; hence, there is an incentive for complete and accurate data (76, 145). Medicaid paid claims also include information on services and treatment received outside of North Carolina that were reimbursed by Medicaid, which provided a more comprehensive analysis of timeliness of services (176).

Some results must be interpreted with caution such as with "Other" race/ethnicity and "noncore areas not adjacent" with place of residence. These data had small cell sizes, leading to wide confidence intervals and imprecise OR.

Despite these limitations, this study had several strengths, including the use of a population-based birth defects registry and GIS. This study used a population-based birth defects surveillance system to verify birth defect diagnoses and demonstrates how useful

such surveillance systems can be in analyzing timeliness of services for children with OFC and children with other birth defects.

This study is the first of its kind in several aspects such as using GIS analysis and examining cleft type and presence of other anomalies. Geographic information systems methods were used to evaluate the role travel distance may play as a potential barrier to accessing care among children with OFC. This study is also unique in that it examines the effect of cleft type, presence of other anomalies, LBW, and preterm birth have on receipt of services. It is important to evaluate specialized service use by these variables because receipt and timeliness of services may vary depending on the condition examined. To date, no study has utilized these variables when examining receipt of services.

This is the first study to examine the timeliness of services such as primary cleft surgery and speech and language therapy per the ACPA recommendations and is the first to do so by cleft type and presence of other anomalies. To date, it is the first study to assess the effect of maternal, child, and system characteristics on timely receipt of primary cleft surgery. This is important to better understand which families of children with OFC are experiencing barriers to accessing services. Having access and receiving timely services can ultimately improve the health outcomes of this population.

Public Health Significance and Opportunities

This study provides unprecedented data on the proportion of children with OFC receiving primary cleft surgery and ancillary services. This study also underscores the importance of assessing different maternal, child, and system characteristics such as maternal age and race, cleft type, presence of other anomalies, and geographical variables to determine their effect

on receipt of medical and ancillary services for children with OFC. These data can identify populations in need of services to better target resources. This is particularly important as children with OFC are at increased risk for learning disabilities and hearing loss, and timely receipt of speech and language therapy and audiology services are critical to their reaching their full potential. Understanding the nature of receipt of timely services among children with OFC are also important for health planning efforts and delivery of services by public agencies such as federal, state, and local Title V CSHCN programs (95). This is essential for allocation of resources such as state and federal Medicaid monies and having sufficient number of health care providers such as in dental and orthodontic professions specifically trained to care for this population. Such information can be used by the North Carolina Commission on Children with Special Health Care Needs, which was mandated by state law to make recommendations for modifications or additions to rules regarding service provision and delivery among CSHCN.

Currently, in North Carolina, and like in most states, state and federal programs exist under the Maternal and Child Health Bureau that will pay for services for CSHCN, including children with OFC. However, these services may include "medically necessary" or "all necessary" treatment contingent on the state law. Consequently, services and treatment may be limited to the length or extent of care in certain states (210, 212). For example, it could exclude dental, scar revision, jaw surgery, and orthodontic procedures and limit the number of speech and language visits per year (210). In North Carolina, these services can be restricted in consultation with the North Carolina Commission on Children with Special Health Care Needs. Federal and state laws also exist that mandate coverage of reconstructive surgical procedures for children with OFC. Like the Maternal and Child Health Bureau

programs, these laws can be restrictive in that they only provide medical services and not ancillary services (210). The exception to this is that federal and state laws were passed mandating schools provide services to CSHCN, which includes speech therapy for children with OFC (210). However, school does not start until age five and for children with OFC, speech therapy is needed much earlier than age five. Both medical and ancillary services are imperative to the overall health and development of these children.

Services and treatment such as hearing, dental, and other specialized services for children with OFC can also be rendered at well-child care visits or the EPSDT benefit, which is the package of Medicaid benefits for children. These visits include comprehensive health and developmental assessments, hearing, vision, and dental services for children birth to 21 years of age and ultimately provide early identification of conditions that can impede children's development (171-173). In addition, through EPSDT children on Medicaid usually have more comprehensive coverage than a usual private health insurance plan, thereby increasing access to needed services to improve children's quality of life. CSHCN like children with OFC need more specialized services such as speech, physical, and occupational therapy, and mental health, which are covered through the EPSDT (171, 173). The low use of specialized services may be due to parents lack of knowledge such services are an entitlement for their children. In addition, it may be due to low provider participation in Medicaid as with dental care and/or inappropriate service denials due to managed care contracts (169, 173-175). Nevertheless, through state law and coverage of specialized services through EPSDT, all children with OFC should be receiving medical and ancillary services. Results from this study prove otherwise, which has grave implications for the health outcomes for these children.

These results are also important when determining reimbursement rates for Medicaid and reauthorization of the State Children's Health Insurance Plan. As indicated, one of the reasons for the shortage of health care professionals specifically trained to treat children with OFC is due to low reimbursement rates. Reimbursement rates and coverage are highly variable with state-funded programs like Medicaid. In some cases, children have been denied coverage, services and treatment because of eligibility criteria and/or not medically necessary. In addition, third party payors have refused to pay for oral health services even if they are associated with the OFC (130, 208). Having insurance, including continuous coverage, and access to services, for both medical and ancillary services for children with OFC are imperative to their overall health and psychosocial well-being and ability to fully function in life.

These data also provide opportunities for craniofacial centers and teams, birth defects surveillance programs, hospitals, and local, state, and national programs and organizations to work collaboratively to increase access to care for children with OFC. Service coordination for CSHCN like children with OFC is an entitlement for eligible children under the United States Individuals with Disabilities Education Act and a mandate under Title V. State-based care coordination programs for children with OFC in North Carolina include Early Intervention/ Infant-Toddler Program for children birth to three, the PreSchool program for children three to five, the Child Service Coordination Program for children from birth to age three who have specific parental/family, neonatal, post-neonatal or diagnosed conditions and children age three to five with at least one of several diagnosed conditions, such as a congenital anomaly. National organizations such as the ACPA, Cleft Palate Foundation, and Family Voices provide opportunities for birth defects programs and craniofacial teams to

collaborate and partner together to improve the lives of children with OFC. Members from these programs can help create and evaluate referral systems to help families of children with OFC obtain the resources and services they need. They can also inform and ensure families that their children are eligible for certain services and that families have the information they need to make informed decisions about their child's care. Decisions include receiving speech and language therapy and audiological services in a timely fashion to improve their child's health.

Because the billing procedures and codes used for these services may be what is at issue and not that children did not receive services, future research should examine the service types and frequency covered by Medicaid and the billing procedures for Medicaid for children with OFC. Additional research should examine other payors of services for children with OFC and discern generic current procedural terminology codes to have more accurate data on children receiving and not receiving timely services. Such research could be conducted by taking a random sample of children with OFC identified by the NCBDMP and determine which children received services and treatment at the various craniofacial centers and teams in the state. Then, these data could be compared to the Medicaid paid claims to identify any discrepancies in billing to determine if the method and codes used for billing were problematic. Additional research should also include the location of services received to determine if any geographical barriers such as distance to craniofacial centers or teams exist and examine actual barriers to accessing care for this population.

Future research should also include an assessment of health outcomes of children with OFC by insurance type to determine appropriate benefit packages and efficient payment mechanisms for both private and public health insurance systems (95). Current federal and

state laws mandating coverage for children with OFC need to be reassessed to ensure all the medical and ancillary service needs of children with OFC are being met.

Conclusion

This study provides unique data on timeliness of services for children with OFC according to the ACPA guidelines. Studies such as this one can help characterize populations of children with OFC who are not receiving services and treatment within a timely manner. These data are important for service and program planning and policy development. Efforts must be made to increase timely receipt of services for infants and children with OFC to improve their health outcomes.

Table 3.1. Summary of the 2000 American Cleft Palate-Craniofacial Association recommended services and treatment for children with orofacial clefts

Type of Service	Timing / Frequency
Primary cleft/lip surgical repair	Within 18 months of life
Audiological assessment	First assessment within first year of life and at least once per year thereafter
Speech and language pathology, including laryngeal function	At least once per year until age four
Otolaryngologic care	Within first six months of life
Dental care, including primary care, routine maintenance, and orthodontic care	At least once per year throughout lifetime
Genetic screening, including follow-up evaluations	Within first two years of life and until puberty for some children
Psychological and social services, including screening evaluations	Periodic through adolescence

Table 3.2. Selected characteristics of children with orofacial clefts by cleft type during the first year of life in North Carolina, 1995-2002[a]

Characteristics	CL[b] N=108 (%)	CP[b] N=210 (%)	CLP[b] N=247 (%)	All OFC[b] N=565 (%)	P-value
Maternal					
Age					
≤ 20 years old	42 (38.9)	48 (22.9)	83 (33.6)	73 (30.6)	0.01*
21-24 years old	32 (29.6)	63 (30.0)	71 (28.7)	166 (29.4)	
25-29 years old	22 (20.4)	56 (26.7)	63 (25.5)	141 (25.0)	
≥ 30 years old	12 (11.1)	43 (20.5)	30 (12.2)	85 (15.0)	
Education					
< High School	44 (40.7)	80 (38.1)	123 (49.8)	247 (43.7)	0.04*
High School	49 (45.4)	85 (40.5)	79 (32.0)	213 (37.7)	
> High School	15 (13.9)	45 (21.4)	45 (18.2)	105 (18.6)	
Race					
White/Non-Hispanic	70 (64.8)	124 (59.1)	154 (62.4)	348 (61.6)	0.62
Black/Non-Hispanic	23 (21.3)	51 (24.3)	52 (21.1)	126 (22.3)	
Hispanic	9 (8.3)	26 (12.4)	34 (13.8)	69 (12.2)	
Other[c]	6 (5.6)	9 (4.3)	7 (2.8)	22 (3.9)	
Number of Living Children					
0	49 (45.4)	84 (40.0)	116 (47.0)	249 (44.1)	0.11
1	38 (35.2)	66 (31.4)	61 (24.7)	65 (29.2)	
≥ 2	21 (19.4)	60 (28.6)	70 (28.3)	151 (26.7)	
Marital Status					
Married	43 (39.8)	105 (50.0)	111 (44.9)	259 (45.8)	0.21
Not married	65 (60.2)	105 (50.0)	136 (55.1)	306 (54.2)	
Initiation of Prenatal Care during 1st Trimester					
Yes	77 (71.3)	169 (80.5)	189 (76.5)	435 (77.0)	0.20
No	31 (28.7)	41 (19.5)	56 (22.7)	128 (22.7)	
Child					
Birthweight					
< 2,500 grams	10 (9.3)	31 (14.8)	41 (16.6)	82 (14.5)	0.19
≥ 2,500 grams	98 (90.7)	179 (85.2)	206 (83.4)	83 (85.5)	
Preterm Birth					
< 37 weeks	11 (10.2)	29 (13.8)	45 (18.2)	85 (15.0)	0.12
≥ 37 weeks	97 (89.8)	181 (86.2)	202 (81.8)	480 (85.0)	
Gender					
Female	45 (41.7)	110 (52.4)	93 (37.7)	248 (43.9)	0.01*
Male	63 (58.3)	100 (47.6)	154 (62.4)	317 (56.1)	
Presence of Other Anomalies[d]					
Isolated	87 (80.6)	107 (51.0)	174 (70.5)	368 (65.1)	<0.00*
Multiple	21 (19.4)	103 (49.1)	73 (29.6)	197 (34.9)	
System					
Source of Prenatal Care					
Health department	60 (55.6)	139 (66.2)	151 (61.1)	350 (62.0)	0.17
Other	48 (44.4)	71 (33.8)	96 (38.9)	215 (38.1)	
Receipt of Maternity Care Coordination Services					
Yes	51 (47.2)	92 (43.8)	109 (44.1)	252 (44.6)	0.83
No	57 (52.8)	118 (56.2)	138 (55.9)	313 (55.4)	

Characteristics	CL[b] N=108 (%)	CP[b] N=210 (%)	CLP[b] N=247 (%)	All OFC[b] N=565 (%)	P-value
System (cont.)					
Receipt of WIC					
Yes	81 (75.0)	139 (66.2)	167 (67.6)	387 (68.5)	0.26
No	27 (25.0)	71 (33.8)	80 (32.4)	178 (31.5)	
Birth Hospital Level of Care					
Level III	45 (41.7)	92 (43.8)	104 (42.1)	241 (42.7)	0.91
Community	63 (58.3)	118 (56.2)	143 (57.9)	324 (57.4)	
Time Traveled[†]					
≤ 30 minutes	17 (20.0)	44 (26.5)	47 (23.2)	108 (23.8)	0.10
31-60 minutes	39 (45.9)	50 (30.1)	60 (29.6)	149 (32.8)	
61-89 minutes	14 (16.5)	24 (14.5)	33 (16.3)	71 (15.6)	
≥ 90 minutes	15 (17.7)	48 (28.9)	63 (31.0)	126 (27.8)	
Perinatal Care Region					
Northwestern	30 (27.8)	58 (27.6)	55 (22.3)	143 (25.3)	0.44
Southwestern	18 (16.7)	26 (12.4)	52 (21.1)	96 (17.0)	
Northeastern	18 (16.7)	37 (17.6)	37 (15.0)	92 (16.3)	
Southeastern	15 (13.9)	23 (11.0)	36 (14.6)	74 (13.1)	
Eastern	18 (16.7)	41 (19.5)	40 (16.2)	99 (17.5)	
Western	9 (8.3)	25 (11.9)	27 (10.9)	61 (10.8)	
Place of Residence					
Metropolitan	73 (67.6)	140 (66.7)	144 (58.3)	357 (63.2)	0.47
Micropolitan	24 (22.2)	44 (21.0)	66 (26.7)	134 (23.7)	
Noncore areas adjacent to metro area or small town	9 (8.3)	18 (8.6)	25 (10.1)	52 (9.2)	
Noncore areas not adjacent to metro area or small town	2 (1.9)	8 (3.8)	12 (4.9)	22 (3.9)	

[a] All children with OFC who were continuously enrolled in Medicaid during the first two years of life (n=406).
[b] CL = cleft lip only; CP = cleft palate only; CLP = cleft lip with cleft palate; OFC = orofacial clefts
[c] Other includes Native American, Asian/Pacific Islander and other non-White
[d] Isolated anomaly = orofacial cleft diagnosis only; multiple anomalies = orofacial cleft + other birth defect diagnosis
[†] Due to not all children with OFC being geocoded, to examine time traveled, the denominator for children with cleft lip was 85, 166 for children with cleft palate and 203 for children with cleft lip with cleft palate. The denominator for all children with OFC was 454.
* $P < 0.05$

Table 3.3. Timely receipt and average age primary cleft surgery occurred among children with orofacial clefts during the first two years of life in North Carolina, 1995-2002[a]

		Received within ACPA Recommendations[b]		Average Age Surgery Occurred within First 18 Months of Life, in Months (Range)
	N	Yes N (%)	No N (%)	
Cleft Type				
Cleft lip	75	66 (88.0)	9 (12.0)	3 (1, 15)
Cleft palate	150	87 (58.0)	63 (42.0)	10 (0, 17)
Cleft lip with cleft palate	183	164 (89.6)	19 (10.4)	4 (0, 16)
All orofacial clefts	406	317 (78.1)	89 (21.9)	5 (0, 17)
Presence of Other Anomalies[c]				
Isolated	260	217 (83.5)	43 (16.5)	5 (0, 17)
Multiple	146	100 (68.5)	46 (31.5)	6 (1, 16)

[a] All children with OFC who were continuously enrolled in Medicaid during the first two years of life (n=406).
[b] The American Cleft Palate-Craniofacial Association (ACPA) recommends primary cleft repair within the first six months of life for children with cleft lip and within 18 months of life for children with cleft palate with or without cleft lip.
[c] Isolated anomaly = orofacial cleft diagnosis only; multiple anomalies = orofacial cleft + other birth defect diagnosis

Table 3.4. Receipt of specialized services among children with orofacial clefts by cleft type during the first year of life in North Carolina, 1995-2002

Service	CL[a] N=108 (%)	CP[a] N=210 (%)	CLP[a] N=247 (%)	All OFC[a] N=565 (%)
Otolaryngologic care	5 (4.6)	46 (21.9)	62 (25.1)	113 (20.0)
Audiological assessment	16 (14.8)	69 (32.9)	75 (30.4)	160 (28.3)
Speech and language therapy	30 (27.8)	108 (51.4)	151 (61.1)	289 (51.2)
Dental	13 (12.0)	30 (14.3)	47 (19.0)	90 (15.9)
Psychological and social services	13 (12.0)	28 (13.3)	29 (11.7)	70 (12.4)

[a] CL = cleft lip only; CP = cleft palate only; CLP = cleft lip with cleft palate; OFC = orofacial clefts

Table 3.5. Timeliness and receipt of specialized services among children with orofacial clefts during the first two years of life in North Carolina, 1995-2002

	Received within first two years of life (N=406)		Timely[a]		Received in Year 1 and Year 2	
	Yes	No	Yes	No	Yes	No
Service	N (%)	N (%)	N (%)	N (%)	N (%)	N (%)
Otolaryngologic care	101 (24.9)	73 (18.0)	44 (7.8)	289 (51.2)	44 (10.8)	130 (32.0)
Audiological assessment	118 (29.1)	288 (70.9)	160 (28.3)	405 (71.7)	50 (12.3)	356 (87.7)
Speech and language therapy	198 (48.8)	208 (51.2)	289 (51.2)	276 (48.8)	134 (33.0)	272 (67.0)
Dental	67 (16.5)	339 (83.5)	90 (15.9)	475 (84.1)	21 (5.2)	385 (94.8)
Psychological and social services	51 (12.6)	355 (87.4)	70 (12.4)	495 (87.6)	51 (12.6)	355 (87.4)
Genetics	56 (13.8)	2 (0. 5)	56 (13.8)	2 (0.5)	**	**

[a] Timely services as defined by the 2000 ACPA: Otolaryngologic care recommended within the first six months of life, so denominator was n=565. Audiological assessment, speech and language therapy, dental services, and psychological and social services, including screening evaluations, recommended within first 12 months of life, so denominator was N=565. Genetic screening, including follow-up evaluations, recommended within first 24 months of life, so denominator was N=406.

** Not assessed because genetic services recommended at least once within first two years of life

Table 3.6. Logistic regression results for the association between selected characteristics and receipt of primary cleft surgery within 18 months of life among children with orofacial clefts (n=406)[a]

Characteristics	Crude Odds Ratio (95% Confidence Interval)	Adjusted Odds Ratio[†] (95% Confidence Interval)
Maternal		
Age		
≤ 20 years old	1.11 (0.57, 2.19)	0.67 (0.27, 1.62)
21-24 years old	1.00	1.00
25-29 years old	0.66 (0.34, 1.27)	0.64 (0.29, 1.43)
≥ 30 years old	0.34 (0.17, 0.70)*	0.25 (0.10, 0.62)*
Education		
< High school	1.55 (0.79, 3.060	0.82 (0.32, 2.11)
High school	0.91 (0.46, 1.77)	0.56 (0.23, 1.36)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	0.60 (0.35, 1.03)	0.30 (0.14, 0.67)*
Hispanic	0.92 (0.43, 1.98)	0.86 (0.32, 2.33)
Other[b]	0.61 (0.21, 1.81)	0.54 (0.13, 2.35)
Number of Living Children		
0	1.00	1.00
1	0.44 (0.25, 0.77)*	0.56 (0.28, 1.21)
≥ 2	0.56 (0.31, 1.01)	1.23 (0.52, 2.93)
Marital Status		
Married	1.00	1.00
Not married	1.25 (0.78, 2.00)	1.23 (0.62, 2.46)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	1.10 (0.64, 1.91)	0.88 (0.43, 1.80)
Child		
Birthweight		
< 2,500 grams	0.84 (0.44, 1.58)	1.07 (0.36, 3.21)
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	0.67 (0.37, 1.23)	0.56 (0.19, 1.63)
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	1.07 (0.67, 1.72)	0.59 (0.32, 1.10)

Characteristics	Crude Odds Ratio (95% Confidence Interval)	Adjusted Odds Ratio† (95% Confidence Interval)
Child (cont.)		
Cleft Type		
Cleft Lip	1.00	1.00
Cleft Palate	0.15 (0.06, 0.34)*	0.11 (0.04, 0.29)*
Cleft Lip with Cleft Palate	0.92 (0.37, 2.28)	0.90 (0.33, 2.44)
Presence of Other Anomalies[c]		
Isolated	1.00	1.00
Multiple	0.43 (0.27, 0.70)*	0.45 (0.24, 0.84)*
System		
Source of Prenatal Care		
Health department	0.93 (0.58, 1.50)	1.40 (0.70, 2.78)
Other	1.00	1.00
Receipt of Maternity Care Coordination Services		
Yes	1.53 (0.95, 2.48)	2.38 (1.16, 4.89)*
No	1.00	1.00
Receipt of WIC		
Yes	1.37 (0.83, 2.26)	0.86 (0.42, 1.77)
No	1.00	1.00
Birth Hospital Level of Care		
Level III	0.77 (0.48, 1.23)	0.80 (0.39, 1.64)
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	0.63 (0.30, 1.31)	0.34 (0.14, 0.84)*
Northeastern	1.38 (0.58, 3.28)	2.53 (0.86, 7.44)
Southeastern	1.01 (0.43, 2.38)	1.54 (0.53, 4.49)
Eastern	0.59 (0.28, 1.21)	0.86 (0.33, 2.22)
Western	0.55 (0.24, 1.26)	0.77 (0.25, 2.32)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	1.15 (0.63, 2.10)	0.67 (0.30, 1.51)
Noncore adjacent	0.45 (0.22, 0.93)*	0.23 (0.08, 0.64)*
Noncore areas not adjacent	0.92 (0.29, 2.91)	1.14 (0.25, 5.20)

[a] All children were continuously enrolled in Medicaid through age two.
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly = orofacial cleft diagnosis only; multiple anomalies = orofacial cleft + other birth defect diagnosis
† Adjusted for all the covariates in the model
* Statistically significant

CHAPTER IV

CONCLUSION

This dissertation examined patterns and selected individual and system characteristics associated with health service use and cost among children with and without OFC during the first year of life. Patterns and predictors of health service use and cost were also assessed by cleft type and presence of other birth defects for children with OFC (Appendix C). This dissertation also assessed the timeliness of certain specialized services among children with OFC within the first two years of life per the ACPA recommendations.

In Chapter Two, health care service use and costs were categorized into medical, inpatient, outpatient, home health, mental health, dental, well-child care, and total. The effect of selected maternal, child, and system characteristics on each health service use and cost category was examined by using Poisson multivariate regression and two-part modeling. The two-part models included binary logit and ordinary least squares regression. The Poisson multivariate regression results in Appendix A were similar to the two-part modeling results, so only the two-part modeling results for costs were reported in Chapter Two.

In Appendices D-F, this dissertation also examined patterns of health service use and costs among children with and without OFC during the second through five years of life (ages one to four). These appendices also included demographic patterns for these age groups and for cases, by cleft type and presence of other birth defects.

In Chapter Three, timeliness of certain specialized services was examined according to the parameters of care for patients with craniofacial conditions, such as OFC, as set forth by the ACPA. Specifically, the proportion of children with OFC who received primary cleft surgery within 18 months of life by cleft type and by presence of other anomalies and any maternal, child, and system factors associated with timely cleft surgery were determined. The proportion of children who received audiology, speech and language therapy, dental, otolaryngologic care, genetic, psychological services by cleft type per the ACPA guidelines also were determined.

The Andersen's Behavioral Model of Health Service Utilization was used to inform this dissertation research. The Behavioral Model of Health Service Utilization posits that peoples' use of health services are a function of their predisposition to use services, factors which impede or enable their use, and their need for care. The three components that comprise this behavioral model are predisposing, enabling, and need characteristics (140-143). In this dissertation, predisposing factors included most of the maternal and child characteristics. Such characteristics included maternal age and education, marital status, race/ethnicity, number of living children, and child's age, gender, and birthweight. Enabling characteristics included most of the system characteristics, which were PNC source, birth hospital level of care, WIC status, perinatal care region, and place of residence. Need factors included both perceived and evaluated need. Perceived need consisted of MCC services, PNC, and service type such as audiology, speech and language therapy, cleft surgery, and mental health services. Evaluated need included cleft type and presence of other anomalies.

Summary of Results

In Chapter Two, this dissertation concluded that children with OFC used significantly more health services and had significantly greater costs than children without this condition during the first year of life. This was especially true for mental health and home health services. Children with OFC used mental and home health services almost 20 times more than unaffected children. The mean cost per child with OFC for mental and home health services was 37 and 45 times higher respectively compared to a child without OFC. The average total cost per child with OFC was six times the average total cost per child without this condition. In the first year of life, the total cost for children with OFC was $12,792,634 compared to $2,212,839 for children without OFC on Medicaid, which were randomly sampled in a 1:1 ratio.

During the first year of life, children with CP and CLP had similar patterns of service use and cost compared to children with CL. The mean number of total paid claims per child with CP was 77.8 and 80.9 for a child with CLP compared to 54.5 for a child with CL. The total mean cost per child with CL was $7,973 compared to $25,575 for a child with CP and $26,817 for a child with CLP. Children with multiple anomalies had almost twice the mean number of total paid claims per child and had almost four times the total cost per child than a child with isolated OFC during the first year of life.

Maternal, child, and system characteristics associated with number of paid claims (i.e., health service use) and cost varied across the different service and cost categories and among children with OFC and unaffected children. After adjusting for these characteristics, children with OFC whose mothers who were 25 years or older, had less than a high school education, received MCC services, and resided in the southeastern region of the state were significantly

more likely to have outpatient costs than their counterparts. In comparison, these maternal and system characteristics were not positively associated with outpatient costs among children without OFC. Children with OFC who resided in the southwestern, southeastern, and western regions of the state were two to three times more likely to have home health costs compared to children living in the northwestern perinatal care region. Perinatal care region was not positively associated with home health costs among children without OFC (Tables 2.8-2.10 and Appendix B Tables 3 and 4). Among children with OFC who had costs, mothers who were Black/non-Hispanic or Hispanic had about 20% less total costs and mothers of "Other" minority race had 25% greater total costs than mothers who were White/non-Hispanic. Minority mothers were not associated with increased or decreased total health care costs among children without OFC.

However, some characteristics that were associated with higher service use and cost among both groups of children were being born LBW and receipt of MCC. After adjusting for all the characteristics, children with and without OFC who were born LBW had significantly higher medical, inpatient, home health, and total service use than children who were not born LBW. In both groups of children, children living in noncore adjacent and noncore areas not adjacent to a metropolitan or small town had greater mental health service use than children living in metropolitan areas. Also, for both groups of children, medical and total service use was significantly higher among mothers who received MCC services than mothers who did not receive such services (Appendix A Tables 1-6 and Appendix B Tables 1-2). Cleft type and presence of other birth defects were strongly associated with higher service use and cost in all service and cost categories among children with OFC.

Characteristics associated with greater health service use and cost among children with OFC and unaffected children underscores the need for services for these children regardless of having a type of birth defect like OFC. Maternity care coordination is a vital component of services and referral to services for families in both groups of children. Programs like MCC should consider their impact on these children, and results from this study could aid in requesting additional resources to enhance the program and target populations in need of services. Increasing funds for care coordination like with MCC could potentially reduce costs in the long-term. With characteristics that differed among cases and controls in terms of health service and cost, this may affect resource allocation by devoting services and care to populations in need of such services and areas of the state where services may be unavailable such as in the noncore adjacent and not adjacent to a metropolitan area or small town.

Some interesting patterns were observed when health service use and costs results in Chapter Two for children with and without OFC during infancy were compared to children ages one through four (Appendix E Tables 1-4 and Appendix F Tables 1-4). The number of continuously enrolled children in Medicaid per year of life decreased with increasing age; however, the overall demographic proportions between cases and controls remained similar (Appendix D).

Mean number of medical, inpatient, and outpatient paid claims per child decreased with increasing age among children with and without OFC. Mean inpatient and outpatient costs per child with OFC and unaffected child steadily decreased over the age groups. However, the mean medical cost per child with OFC peaked in the third year of life whereas for children without this condition, the mean medical cost per child decreased over the first five years of life (Appendix E Tables 1-4 and Appendix F Tables 1-4). These results for children

with OFC were a bit perplexing because it was thought the highest medical and inpatient service use and costs would have occurred in the second year of life due to about 78% of children receiving primary cleft surgery during this time period. However, the results for children with OFC were most likely due to the numerous surgeries conducted to repair the CL and/or CP during these years. This would contribute to higher inpatient service use and cost among this population.

Even though the utilization ratio between cases and controls for mental health services decreased as children grew older, the mean number of mental health paid claims and mean cost of mental health services per child with OFC increased until age two and then decreased at age three and four. This was a bit surprising because it was expected that mental health services and costs among children with OFC would have increased over time and not decreased due to the increase in mental health needs and psychosocial well-being of children with OFC as a result of their cleft (Appendix E Tables 1-4 and Appendix F Tables 1-4).

Trends in home health service use and cost varied across the age groups among both cases and controls. The mean cost of home health services per child with OFC was highest in the second year of life. During the second year of life, the mean cost of home health services per child with OFC was 88.8 times higher than a child without this condition (Appendix E Tables 1-4 and Appendix F Tables 1-4). As with mental health services and costs, these results were unexpected because it was thought that there would have been a direct linear relationship between increasing age and home health service use and cost among children with OFC.

Mean number of dental paid claims and mean dental cost per child increased with increasing age for both cases and controls, which was to be expected. Dental service and cost ratios between cases and controls were highest in the second year of life. The highest mean

cost for per child for dental services for cases and controls was in the fifth year of life (Appendix E Tables 1-4 and Appendix F Tables 1-4). The results for dental service use and cost were to be expected as many children do not use dental services in infancy and thus have less dental costs in the first year of life.

Overall, children with OFC utilized health services almost twice as much compared to unaffected children during the first five years of life. Over the first five years of life, the mean total cost per child with OFC ranged from six to ten times higher than the mean total cost per unaffected child (Appendix E Tables 1-4 and Appendix F Tables 1-4). The five-year cumulative average cost per child with OFC was $65,319 compared to $9,190 cumulative average cost per child without this condition. The total cumulative cost for children with OFC was $25,706,332 compared to $3,554,222 for unaffected children, which were randomly sampled in a 1:1 ratio.

For all five years of life, the mean total service use and cost per child with CP or CLP were similar compared to a child with CL. The average number of paid claims for home and mental health service use and cost per child with CL were significantly less than for a child with CP or CLP. During the first five years of life, total mean cost per child with CP and CLP were three to seven times more than the mean cost per child with CL (Appendix E Tables 5-6 and Appendix F Tables 5-6). The five-year cumulative cost for children with CL was $1,272,094 compared to $11,120,982 for children with CP and $13,313,256 for children with CLP.

During the first five years of life, children with multiple anomalies utilized services almost twice as much as children with an isolated anomaly. Total mean cost per child with multiple anomalies ranged from four to eight times more than a child with an isolated

anomaly (Appendix E Tables 5-6 and Appendix F Tables 5-6). The five-year cumulative cost for children with multiple anomalies was about 13 million dollars more than children with an isolated anomaly, $19,366,368 and $6,339,964 respectively.

Unfortunately, this dissertation was unable to examine the effects of selected maternal, child, and system characteristics on health service use and cost for ages one through four due to small numbers of continuously enrolled children in Medicaid, especially among children with OFC. Children who were enrolled per year of life were used instead of children who were continuously enrolled all five years of life due to small numbers in the latter category. The cross-sectional analysis of children continuously enrolled each year of life was representative of the longitudinal analysis of children continuously enrolled all five years of life. Determining these factors is critical especially among children with OFC because needs differ at various developmental stages and the services offered through the United States Individuals with Disabilities Education Act and a mandate under Title V can differ according the child's age and needs. For example, dental, orthodontic, and psychological/mental health services and needs are greater as the child increases with age due to the development of speech and language and teeth. In addition, eligibility for early intervention and child service coordination programs differ according to the child's age and needs. Knowing which maternal, child, and system characteristics are associated with underutilization of health services can help identify populations in need of services and target service delivery and resources, including referral of CSHCN to the Early Intervention/Infant Toddler program for children birth to three and the Child Service Coordination Program in North Carolina for children three to five years old.

Findings from Chapter Two and Appendices E and F suggest that costs of care for children with OFC are high and highly variable driven by certain maternal, child, and system characteristics such as cleft type and presence of other anomalies. This research also found which service categories are drivers of cost such as medical and inpatient among children with and without OFC.

From Chapter Three, this dissertation found that the majority of children with OFC had primary cleft surgical repair within 18 months of life, which was within the ACPA guidelines. Of those children who received timely surgery, mean age surgery occurred was five months. The average age primary cleft surgery occurred varied significantly by cleft type, but not by presence of other anomalies. Children whose mothers received MCC services, received PNC at a local health department, or lived in the northeastern and southeastern region of the state were more likely to receive primary cleft surgery within 18 months of life than their counterparts. Results from Chapter Three also suggest that many children with OFC did not receive other necessary specialized services per the ACPA guidelines.

Overview of Strengths and Potential Limitations

This study provides unprecedented and comprehensive information on health service use and costs of children with OFC compared to unaffected children. To date, this study is the only one that determines health service use and cost by several service categories. In addition, it is the only study to examine service use and cost for this population among several age groups. Previous studies on health service use and cost of children with OFC only examined one age group and a few health service categories. Patterns of service use and costs

are important when determining which services are utilized most at different times in life, especially for CSHCN, such as children OFC.

Another strength is that this study stratified health service use and cost by presence of other anomalies and by three different cleft types, which were CL, CP, and CLP. The few previous studies on health service use and costs of children with OFC have grouped OFC into two groups, CP and CL with and without CP. This classification masks the effects of CL and CLP. In fact, this study found that children with CP and CLP were more similar in service use and cost than children with CL. Stratifying by cleft type and presence of other birth defects is important for allocation of resources, targeting care coordination and early intervention, etiologic research, and referral of services. This is the first study to examine health service use and cost of children with OFC by these three cleft types and by presence of other anomalies.

This dissertation is the first to assess the effect of selected maternal, child, and system characteristics on health service use and cost among children with OFC compared to children without this condition. Several child characteristics like being born LBW and preterm and system characteristics such as receipt of PNC and MCC, perinatal care region, and place of residence were examined. These characteristics have not been investigated in previous studies on health service use and cost of CSHCN. Novel techniques such as perinatal care region and Urban Influence Codes for place of residence were utilized as geographical variables to determine their association with health service use and cost among cases and controls. Perinatal care region is important for current planning of Title X service delivery and program development within the state. In addition, the regions are a useful way to look at maternal and child health indicators because they are population-based (148, 149). Urban

Influence Codes are helpful in distinguishing the effects of adjacency compared to size of the area on access and health service utilization (152). Urban Influence Codes are also beneficial when examining the structure of health care systems and whether outcomes or care are possibly related to the complexity of the medical or health community (150, 151). This is important when examining access to health care and service utilization because some previous research in health services have defined place of residence as urban or rural. This classification masks any geographical differences and does not consider heterogeneity within counties. Including these variables can assist in targeting resources and services to populations in need of services such as in the southeastern and western perinatal care regions and in noncore areas adjacent and not adjacent to metropolitan areas. By including these geographical variables, service program planners and public health practitioners can have a better understanding of which populations among children with and without OFC are underutilizing services. Additionally, they can better understand where access to services may need to be increased in certain areas of the state. Increasing health care provider availability such as with craniofacial centers and teams in the western and southeastern regions of the state can help ensure children have access to and receive services, especially services entitled under Medicaid such as with the EPSDT services.

Another strength of this study included employing GIS analysis. This study is the first to use GIS methods to evaluate the role travel distance may play as a potential barrier to accessing care such as at craniofacial centers and teams among children with OFC. In this study, time traveled did not contribute significantly to the model predicting receipt of timely cleft surgery. Future studies should include this variable to determine whether it is a barrier to care for this population. In addition, this is the first study to examine the effect of time

traveled after adjusting for important confounding variables such as maternal age and race, birth hospital level of care, receipt of PNC, WIC, and MCC, and geographic variables such as perinatal care region and place of residence.

Some potential limitations with the GIS analysis and the use of the maternal residential address at birth need to be acknowledged. Families may have moved during the first year of life, which could have decreased or increased travel time to the nearest craniofacial center or team. In addition, children may not have received services and treatment at the closest craniofacial center or team to their residence. Children may receive their primary craniofacial care at multiple craniofacial centers or teams or at hospitals or Children's Developmental Service Agencies in their local communities. Thus, the time traveled results may have underestimated the true distance and time traveled to receive craniofacial care. This may have been why the time traveled variable was not a significant predictor of timely cleft surgery.

Other strengths included employing a population-based birth defects registry and examining actual costs to Medicaid. This study used a population-based birth defects surveillance system to verify birth defect diagnoses whereas previous studies on health service use and cost that utilized Medicaid diagnoses codes did not verify the condition. This study also examined actual costs reimbursed by Medicaid to better understand the direct financial burden of families affected by OFC.

Lastly, this study is the first study to examine timeliness of services per nationally recommended guidelines as set forth by the ACPA. Receiving timely services and treatment is especially important for children with OFC to help prevent hearing loss, speech impairments, learning disabilities, and other secondary conditions.

Despite these strengths, several potential limitations existed with this research, such as exclusion of possible effect measure modifiers and confounding variables and limited generalizability of the results. Cleft laterality, a potential effect measure modifier, was unable to be determined because data were unavailable. This is important because children with bilateral clefts usually require additional surgeries and specialized services, thereby increasing service use and cost for these children. Potentially confounding variables that were also unavailable for this study included other children with birth defects and/or other children with OFC within the same family, maternal or paternal occupation, and previous service participation or knowledge of MCC, WIC, Medicaid, and PNC. All of these covariates could have contributed to health service use, cost, and the timeliness of services.

Generalizability of the findings may be limited to low-income children with and without OFC in North Carolina. This is because this study represents only a subpopulation of children in the state and only one type of health insurance, and thus may not be representative of all children with OFC and unaffected children. Using Medicaid data was the only mechanism to track health service use of this population in the state.

Restricting the analysis to children continuously enrolled in Medicaid was also strength for several reasons. One, the majority of service use and costs were most likely captured because services received out of state that were reimbursed by Medicaid were included. Two, the potential for confounding by socioeconomic status was reduced.

Other potential limitations included small cell sizes and examining only direct health care costs. Some results should be interpreted with caution such as with "Other" race/ethnicity and "noncore areas" with place of residence due to small cell sizes, especially among children with OFC. This study only included crude direct health care costs and thus excluded

other costs for children with OFC such as out-of-pocket, caregiver, and psychosocial costs. In addition, Medicaid usually pays "a smaller proportion of the amount billed by providers than other third-party payers" (213). Hence, this study represents only a portion of the total costs for children with OFC and most likely underestimates the true cost of children with OFC during the first five years of life.

Lastly, unfortunately, this study was unable to determine characteristics associated with health service use and cost for children ages one through four. Too few children were continuously enrolled in these different age groups, especially for children with OFC. Characteristics associated with higher service use and cost would most likely be different for the various age groups due to parental perceptions of need and developmental and medical needs of the child in both groups of children.

Public Health Significance

Service use, costs, and timeliness of services among children with OFC was recently identified by the CDC as priority research areas due to the high cost of children with birth defects like OFC (33). Understanding the nature of health care service utilization, costs, and timeliness of services among children with OFC are important for health planning efforts and service delivery by public agencies such as federal, state, and local Title V CSHCN programs (95). This study indicates that children without OFC whose mothers were less educated, younger and older than 21-24 years of age, and of minority/ethnic origin, and families living in certain areas of the state like the western region and noncore areas adjacent and not adjacent to metropolitan areas are less likely to use specific health services such as home health and mental health. This is probably associated with lack of access to such services

and/or lack of awareness or knowledge these families are eligible for such services (180-182).

This study substantiates previous studies on CSHCN that found higher rates of mental health and dental unmet needs among CSHCN. As such, this study underscores the importance for resources and trained health care providers, especially in mental health and dental services, for families of children with OFC. This study also indicates that receipt of certain services such as initiation of PNC in the first trimester, WIC, and MCC can be positively or negatively associated with outpatient, mental health, home health, and total service use and costs in children with and without OFC. This is important because maternity outreach workers, community transition coordinators and case managers can utilize this information to link families and children to services and ensure a referral system is in place for CSHCN such as children with OFC. It is also important because promoting and establishing referral systems to craniofacial rehabilitative teams and conducting craniofacial health surveillance among children with craniofacial anomalies like OFC is part of *Healthy People 2010* objectives 21-15 and 21-16 (139).

Differences in service use and cost by place of residence, perinatal care region, and maternal race/ethnicity, age, and education among children with OFC should be considered in organizing needs for this population in the state. In addition, population-based information about children with OFC and their families is important for assessing needs and evaluating pediatric initiatives at the state level. Determining factors that mediate Medicaid costs can guide future investments in programs that coordinate care or otherwise serve families of children affected by OFC.

This study also underscores the importance of the goals and roles of public health, including proper identification and referral of children with OFC. Studies such as this one can help identify and refer certain families of children with OFC for early intervention as it is well known that early intervention has a positive impact on CSHCN (187-190). In addition, this dissertation research demonstrates the importance of collaborations and partnerships between birth defects registries, community hospitals, local community services, craniofacial centers and teams, and families of children with OFC. It is through these collaborations and partnerships that all children with OFC can be referred to services and will receive timely medical and ancillary services per the ACPA recommendations.

This study provides information on patterns and predictors of health care service utilization and costs that may be applied in determining appropriate benefit packages and efficient payment mechanisms for both private and public health insurance systems (95). Specifically, this information can be used to identify: populations in need of services; availability of services; and categories of high and low utilization and cost. Results from the timeliness of services among children with OFC can be used by the North Carolina Commission on Children with Special Health Care Needs, which makes recommendations for modifications or additions to rules regarding service provision and delivery among CSHCN.

Lastly, this study underscores the importance of continued health insurance and adequate coverage for CSHCN such as children with birth defects to have access to the services they need to improve the overall health of these children. This study indicates that despite coverage of services through Medicaid and services provided through the EPSDT program, many children with OFC may still not be receiving necessary recommended specialized

services and may not be having all their necessary services covered through insurance. This may be due to restrictions on provider referrals, number of visits, pre- and post-orthodontic appliances for surgery and/or number of surgeries for children with OFC. The current coverage via Medicaid may not be adequate to meet the needs of this population, which can place additional burdens on families to pay directly for care.

Recommendations for Next Steps

This dissertation provides useful and current information on patterns and predictors of health service use and cost among children with and without OFC. It also provides data on the timeliness of services among children with OFC. The work completed thus far provides opportunities to pursue additional analyses that can address important policy-related questions for children with birth defects. Conducting further analyses of existing data could assist the Maternal and Child Health Bureau in accomplishing its goals for the *2010 Healthy People* objectives and address research priorities areas for children with OFC as identified by CDC. Future research could also assist in increasing the number of medical homes and referral systems for CSHCN like children with OFC. Additional analyses could inform national and state commissions on CSHCN and provide data for national and state legislators in hopes of changing current health insurance policies for CSHCN, especially for children with OFC. In addition, further analyses can give families and policy makers a better depiction of the true financial burden of children with OFC. Recommendations for some specific research questions and subsequent studies are outlined below. This is not meant to be an exhaustive list and some studies could be combined into one study. These research questions are ranked in order of priority according to the author.

- What services are rendered for generic procedural codes currently utilized by craniofacial centers and teams in other states for reimbursement by Medicaid?

Because craniofacial centers and teams may use different procedural codes for a variety of services, including codes that are utilized by craniofacial centers in other states may be warranted. Additional data from Medicaid paid claims such as provider specialty codes and provider location could be employed to discern specific services rendered to children with OFC. For this dissertation, only certain variables from Medicaid paid claims were available. Using both type of information would most likely identify more children who received services per the ACPA guidelines. To determine the extent this may have affected results in this dissertation, a sensitivity analysis could be conducted by matching a random sample of children with OFC in this study with children seen at craniofacial centers or teams in the state.

- Who are the other payors for services and treatment and what are the costs for these services and treatment among children with OFC?

This research question could be answered by matching the data in this dissertation to private health insurance companies, craniofacial centers, and/or hospital such as St. Jude's Children's Hospital to determine which children received services, the type of services received, and the costs of such services. These analyses would directly address one of CDC's research priority areas for children with OFC and would be of substantial interest to consumer groups.

- What are the barriers to care, including eligibility requirements for private and public health insurance, eligibility for services, and distance traveled to health care providers

among children with OFC? Moreover, are these barriers affecting receipt of timely services among this population?

To address this question, data on the location of services actually received among children with OFC would need to be determined. This information could be obtained using additional provider codes in Medicaid and/or asking parents of children with OFC where their child received primary craniofacial services. In addition, current residential addresses would need to be obtained to use in geocoding to better depict the distance traveled to receive craniofacial services. A survey assessing barriers to care such as perceived need of services, perceived eligibility for health care benefits or services, enrollment procedures for health insurance, and distance to care could be administered to parents of children with OFC.

- What are the out-of-pocket costs such as caregiver costs and indirect costs to families of children with OFC?

Findings from the dissertation confirm children with OFC cost significantly more than unaffected children. However, this study only included direct medical costs. To assess direct costs not paid by Medicaid, indirect costs, and non-medical costs, surveys could be administered to parents of children with OFC asking about out-of-pocket costs. These families should be identified by birth defects surveillance programs and/or craniofacial centers so the diagnoses of OFC are verified. Updated cost information from different types of health insurance matched with birth defects registries and vital statistics could assess mortality and morbidity costs of children with OFC.

- What are the relationships between coordinated systems of care like with craniofacial teams, medical homes, and physical and mental health services for children with OFC?

An important national policy objective is encouraging the development and delivery of coordinated care like with craniofacial teams and medical homes. Another objective is to coordinate physical and behavioral health services to manage complex chronic conditions, such as children with birth defects, in low-income populations (81). Development of these systems were recently posited by the American Academy of Pediatrics and the National Center of Medical Home Initiatives for Children with Special Health Care Needs (8, 115, 116). To accomplish these research objectives, more information is needed on the relationship between the use of medical and mental health services. Additional analysis is needed on whether children who use medical services are more likely to utilize mental and home health services and more likely to be users of specialty medical care. Data from this dissertation could be used to address this question.

- What are the patterns and predictors of service use and cost over the lifespan of children with OFC, not just the first five years of life?

To assess this question, longitudinal cohort studies of children with OFC from birth throughout the child's life would need to be conducted. Such research would most likely require matching of several administrative data from birth defects programs, service programs, hospitals, craniofacial centers and various health insurance companies to determine the types and timing of services. Specific services and costs would need to be identified. Longitudinal cohort studies are also needed to determine any further trends in untimely receipt of services throughout the lifetime of children with OFC. As discussed in this dissertation, service use and costs can vary according to the child's needs and age.

- What are the effects of insurance type on children with OFC receiving timely services?

The longitudinal study previously mentioned could also examine this question along with health outcomes of children with OFC to determine if certain benefit packages and mechanisms are better than others for children with OFC. Results from this study could inform families and federal and state legislators about health insurance policies and perhaps underscore the need to reassess such policies.

In sum, this study provides unique and unprecedented data on health service use, cost, and timeliness of services for children with OFC. It confirms that children with OFC significantly utilize services and cost significantly more than unaffected children. This dissertation indicates that children with CP and CLP have similar service utilization and cost patterns than children with CL. Previous studies did not consider CL alone and CLP separately. As expected, these results demonstrate that children with multiple anomalies utilize services and cost significantly more than children with OFC alone. Previous studies did not evaluate the presence of other anomalies, which is important in determining individualized treatment plans and the timing and receipt of services such as primary cleft surgery. Studies such as this one can help identify children with OFC who are not receiving services and treatment within a timely manner. Results presented in this dissertation are important for service and program planning and policy development. Efforts must be made to increase timely receipt of services for infants and children with OFC to improve their health outcomes. Results from this dissertation should ultimately improve access to services and, more importantly, the overall health and development of children with OFC in North Carolina and the United States.

APPENDIX A: POISSION REGRESSION RESULTS FOR PREDICTORS OF HEALTH SERVICE USE AMONG CHILDREN WITH AND WITHOUT OROFACIAL CLEFTS DURING THE FIRST YEAR OF LIFE IN NORTH CAROLINA, 1995-2002

Table 1. Poisson Regression of Selected Characteristics on the Number of Medical Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=5,638)** Count Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	0.94 (0.80, 1.10)	1.00 (0.97, 1.03)
21-24 years old	1.00	1.00
25-29 years old	0.98 (0.84, 1.14)	0.97 (0.94, 1.01)
≥ 30 years old	0.90 (0.75, 1.09)	1.01 (0.97, 1.05)
Education		
< High school	1.08 (0.91, 1.29)	0.99 (0.95, 1.02)
High school	1.07 (0.91, 1.27)	0.98 (0.95, 1.01)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	1.02 (0.87, 1.19)	0.94 (0.92, 0.97)*
Hispanic	0.86 (0.70, 1.05)	0.92 (0.88, 0.95)*
Other[b]	1.16 (0.88, 1.54)	0.96 (0.90, 1.03)
Number of Living Children		
0	1.00	1.00
1	1.05 (0.91, 1.21)	0.98 (0.95, 1.01)
≥ 2	0.97 (0.82, 1.14)	0.99 (0.95, 1.02)
Marital Status		
Married	1.00	1.00
Not married	0.91 (0.80, 1.03)	1.03 (1.00, 1.06)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	0.93 (0.80, 1.07)	0.92 (0.89, 0.95)*
Child		
Birthweight		
< 2,500 grams	1.31 (1.07, 1.60)*	1.17 (1.11, 1.23)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.16 (0.95, 1.41)	1.17 (1.12, 1.22)*
≥ 37 weeks	1.00	1.00

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
Child (cont.)		
Gender		
Female	1.00	1.00
Male	1.05 (0.93, 1.18)	1.07 (1.05, 1.10)[*]
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.93 (0.81, 1.07)	0.96 (0.94, 0.99)[*]
Receipt of Maternity Care Coordination Services		
Yes	1.20 (1.05, 1.38)[*]	1.06 (1.03, 1.09)[*]
No	1.00	1.00
Receipt of WIC		
Yes	0.86 (0.74, 0.98)[*]	1.04 (1.01, 1.07)[*]
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.10 (0.96, 1.26)	0.94 (0.91, 0.96)[*]
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	1.04 (0.86, 1.26)	0.81 (0.78, 0.84)[*]
Northeastern	1.35 (1.12, 1.62)[*]	1.09 (1.05, 1.13)[*]
Southeastern	1.25 (1.02, 1.55)[*]	0.96 (0.93, 1.00)
Eastern	1.14 (0.94, 1.38)	1.03 (0.99, 1.07)
Western	1.31 (1.07, 1.62)[*]	1.21 (1.16, 1.26)[*]
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	1.00 (0.85, 1.17)	0.95 (0.93, 0.98)[*]
Noncore adjacent	0.95 (0.77, 1.17)	0.90 (0.86, 0.94)[*]
Noncore areas not adjacent	0.95 (0.68, 1.32)	0.88 (0.83, 0.94)[*]

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 2. Poisson Regression of Selected Characteristics on the Number of Inpatient Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=5,638)** Count Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	0.99 (0.84, 1.15)	0.99 (0.96, 1.03)
21-24 years old	1.00	1.00
25-29 years old	0.97 (0.84, 1.13)	0.99 (0.95, 1.02)
≥ 30 years old	0.85 (0.70, 1.02)	0.97 (0.93, 1.01)
Education		
< High school	1.10 (0.92, 1.30)	1.07 (1.03, 1.11)*
High school	1.04 (0.89, 1.23)	1.01 (0.98, 1.05)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	0.94 (0.80, 1.10)	1.00 (0.97, 1.03)
Hispanic	0.83 (0.69, 1.01)	0.97 (0.93, 1.01)
Other[b]	1.27 (0.97, 1.65)	0.98 (0.92, 1.05)
Number of Living Children		
0	1.00	1.00
1	0.97 (0.84, 1.11)	1.02 (0.99, 1.05)
≥ 2	1.02 (0.87, 1.20)	1.01 (0.98, 1.05)
Marital Status		
Married	1.00	1.00
Not married	0.85 (0.75, 0.96)*	1.00 (0.97, 1.03)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	0.96 (0.83, 1.10)	0.96 (0.93, 0.99)
Child		
Birthweight		
< 2,500 grams	1.26 (1.03, 1.53)*	1.16 (1.10, 1.21)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.09 (0.89, 1.33)	1.14 (1.09, 1.20)*
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	0.96 (0.86, 1.08)	1.07 (1.04, 1.10)*

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.86 (0.75, 0.98)[*]	1.00 (0.97, 1.03)[*]
Receipt of Maternity Care Coordination Services		
Yes	1.02 (0.90, 1.17)	1.01 (0.98, 1.04)
No	1.00	1.00
Receipt of WIC		
Yes	0.94 (0.82, 1.08)	1.03 (1.00, 1.06)
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.11 (0.97, 1.26)	0.98 (0.95, 1.01)
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	0.96 (0.81, 1.15)	1.00 (0.96, 1.04)
Northeastern	0.97 (0.80, 1.17)	0.98 (0.94, 1.02)
Southeastern	1.10 (0.90, 1.35)	1.05 (1.01, 1.09)[*]
Eastern	1.11 (0.93, 1.34)	0.98 (0.95, 1.02)
Western	1.25 (1.02, 1.52)[*]	0.97 (0.93, 1.02)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	1.06 (0.91, 1.23)	1.00 (0.97, 1.03)
Noncore adjacent	1.03 (0.84, 1.26)	1.02 (0.98, 1.07)
Noncore areas not adjacent	1.14 (0.85, 1.52)	0.99 (0.93, 1.06)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 3. Poisson Regression of Selected Characteristics on the Number of Outpatient Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=5,638)** Count Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	0.82 (0.66, 1.02)	1.04 (0.94, 1.14)
21-24 years old	1.00	1.00
25-29 years old	0.91 (0.74, 1.12)	0.97 (0.87, 1.07)
≥ 30 years old	0.94 (0.73, 1.21)	0.94 (0.84, 1.06)
Education		
< High school	1.23 (0.96, 1.56)	1.22 (1.09, 1.36)*
High school	1.19 (0.95, 1.50)	1.11 (1.00, 1.23)*
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	0.96 (0.77, 1.20)	1.11 (1.01, 1.21)*
Hispanic	0.99 (0.77, 1.27)	1.16 (1.03, 1.30)*
Other[b]	1.56 (1.10, 2.22)*	1.04 (0.85, 1.26)
Number of Living Children		
0	1.00	1.00
1	0.95 (0.78, 1.14)	0.96 (0.88, 1.05)
≥ 2	0.85 (0.68, 1.05)	0.91 (0.82, 1.02)
Marital Status		
Married	1.00	1.00
Not married	0.95 (0.80, 1.14)	1.06 (0.98, 1.15)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	0.90 (0.74, 1.09)	0.81 (0.75, 0.89)*
Child		
Birthweight		
< 2,500 grams	1.26 (0.96, 1.65)	1.17 (1.02, 1.35)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.15 (0.88, 1.51)	1.26 (1.11, 1.43)*
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	1.09 (0.93, 1.27)	1.25 (1.16, 1.34)*

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.96 (0.80, 1.16)	1.09 (1.01, 1.18)[*]
Receipt of Maternity Care Coordination Services		
Yes	1.09 (0.91, 1.31)	1.06 (0.97, 1.14)
No	1.00	1.00
Receipt of WIC		
Yes	0.98 (0.81, 1.19)	1.14 (1.05, 1.25)[*]
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.23 (1.03, 1.48)[*]	1.26 (1.16, 1.37)[*]
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	0.96 (0.74, 1.23)	1.08 (0.96, 1.21)
Northeastern	1.24 (0.98, 1.57)	1.02 (0.92, 1.15)
Southeastern	1.12 (0.85, 1.48)	0.97 (0.86, 1.09)
Eastern	0.94 (0.72, 1.23)	0.73 (0.64, 0.83)
Western	0.97 (0.72, 1.30)	1.07 (0.94, 1.22)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	0.87 (0.71, 1.08)	0.97 (0.88, 1.07)
Noncore adjacent	0.87 (0.64, 1.17)	1.33 (1.18, 1.51)[*]
Noncore areas not adjacent	0.90 (0.55, 1.47)	1.39 (1.15, 1.68)[*]

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 4. Poisson Regression of Selected Characteristics on the Number of Mental Health Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=5,638)** Count Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	1.00 (0.55, 1.83)	0.38 (0.20, 0.73)[*]
21-24 years old	1.00	1.00
25-29 years old	1.05 (0.60, 1.85)	0.81 (0.44, 1.52)
≥ 30 years old	0.77 (0.36, 1.68)	0.78 (0.39, 1.58)
Education		
< High school	1.45 (0.72, 2.94)	1.72 (0.88, 3.39)
High school	1.12 (0.56, 2.22)	0.81 (0.41, 1.62)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	0.95 (0.50, 1.79)	0.92 (0.51, 1.64)
Hispanic	1.59 (0.83, 3.03)	1.04 (0.48, 2.25)
Other[b]	2.31 (0.91, 5.89)	0.18 (0.01, 2.72)
Number of Living Children		
0	1.00	1.00
1	1.01 (0.58, 1.73)	0.79 (0.44, 1.43)
≥ 2	0.94 (0.51, 1.73)	0.86 (0.45, 1.63)
Marital Status		
Married	1.00	1.00
Not married	0.92 (0.57, 1.48)	1.51 (0.88, 2.60)
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	1.78 (1.09, 2.90)[*]	1.14 (0.68, 1.91)
Child		
Birthweight		
< 2,500 grams	1.25 (0.65, 2.39)	4.62 (2.34, 9.12)[*]
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	2.67 (1.41, 5.06)[*]	1.98 (0.99, 3.96)
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	1.38 (0.88, 2.18)	2.00 (1.24, 3.24)[*]

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	1.16 (0.70, 1.92)	0.96 (0.58, 1.59)
Receipt of Maternity Care Coordination Services		
Yes	0.88 (0.53, 1.45)	1.61 (0.94, 2.76)
No	1.00	1.00
Receipt of WIC		
Yes	1.18 (0.69, 2.01)	0.62 (0.36, 1.06)
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.01 (0.61, 1.69)	1.29 (0.76, 2.21)
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	0.62 (0.31, 1.21)	0.38 (0.14, 1.02)
Northeastern	0.40 (0.19, 0.85)[*]	0.35 (0.14, 0.83)[*]
Southeastern	0.49 (0.23, 1.07)	0.60 (0.28, 1.30)
Eastern	0.68 (0.34, 1.36)	0.83 (0.40, 1.70)
Western	0.60 (0.27, 1.31)	1.59 (0.81, 3.15)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	0.86 (0.47, 1.58)	1.33 (0.73, 2.41)
Noncore adjacent	2.20 (1.11, 4.34)	1.52 (0.74, 3.12)
Noncore areas not adjacent	2.16 (0.76, 6.10)	1.55 (0.54, 4.49)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 5. Poisson Regression of Selected Characteristics on the Number of Home Health Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=5,638)** Count Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	1.17 (0.52, 2.60)	0.66 (0.41, 1.05)
21-24 years old	1.00	1.00
25-29 years old	1.98 (1.03, 3.81)*	1.01 (0.63, 1.61)
≥ 30 years old	0.71 (0.24, 2.05)	0.82 (0.48, 1.40)
Education		
< High school	1.36 (0.57, 3.25)	0.82 (0.52, 1.30)
High school	1.27 (0.57, 2.83)	0.48 (0.30, 0.76)*
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	1.20 (0.58, 2.49)	0.77 (0.52, 1.15)
Hispanic	0.42 (0.13, 1.29)	0.62 (0.31, 1.22)
Other[b]	1.34 (0.41, 4.39)	0.60 (0.23, 1.58)
Number of Living Children		
0	1.00	1.00
1	1.98 (1.07, 3.68)*	0.80 (0.52, 1.22)
≥ 2	0.90 (0.40, 2.01)	1.01 (0.63, 1.62)
Marital Status		
Married	1.00	1.00
Not married	0.68 (0.37, 1.25)	1.64 (1.08, 2.48)*
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	0.57 (0.28, 1.18)	1.26 (0.86, 1.84)
Child		
Birthweight		
< 2,500 grams	3.52 (1.35, 9.17)*	5.54 (3.36, 9.14)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	0.46 (0.16, 1.29)	3.08 (1.86 5.09)*
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	2.24 (1.25, 4.01)*	1.54 (1.09, 2.16)*

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	1.02 (0.55, 1.88)	0.82 (0.57, 1.19)
Receipt of Maternity Care Coordination Services		
Yes	2.20 (1.17, 4.14)[*]	1.34 (0.91, 1.99)
No	1.00	1.00
Receipt of WIC		
Yes	0.40 (0.21, 0.76)[*]	1.12 (0.74, 1.70)
No	1.00	1.00
Birth Hospital Level of Care		
Level III	2.84 (1.53, 5.25)[*]	1.77 (1.19, 2.64)[*]
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	4.93 (1.85, 13.15)[*]	1.54 (0.94, 2.52)
Northeastern	4.57 (1.67, 12.51)[*]	0.32 (0.16, 0.64)[*]
Southeastern	3.49 (1.08, 11.30)[*]	1.46 (0.92, 2.32)
Eastern	2.56 (0.87, 7.55)	0.22 (0.09, 0.54)[*]
Western	1.70 (0.47, 6.12)	1.18 (0.65, 2.14)
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	1.93 (0.97, 3.83)	0.73 (0.45, 1.19)
Noncore adjacent	1.23 (0.45, 3.37)	0.77 (0.40, 1.50)
Noncore areas not adjacent	1.81 (0.32, 10.23)	2.34 (0.84, 6.51)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 6. Poisson Regression of Selected Characteristics on the Number of Total Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)	**Children without Orofacial Clefts (n=5,638)** Count Ratios[†] (95% Confidence Intervals)
Maternal		
Age		
≤ 20 years old	0.94 (0.80, 1.09)	1.00 (0.97, 1.03)
21-24 years old	1.00	1.00
25-29 years old	0.99 (0.86, 1.15)	0.98 (0.95, 1.01)
≥ 30 years old	0.91 (0.76, 1.10)	1.01 (0.97, 1.04)
Education		
< High school	1.11 (0.93, 1.31)	0.99 (0.96, 1.03)
High school	1.09 (0.93, 1.28)	0.98 (0.95, 1.01)
> High school	1.00	1.00
Race/Ethnicity		
White/non-Hispanic	1.00	1.00
Black/non-Hispanic	1.01 (0.87, 1.18)	0.95 (0.92, 0.98)*
Hispanic	0.88 (0.73, 1.06)	0.93 (0.89, 0.96)*
Other[b]	1.22 (0.94, 1.59)	0.96 (0.90, 1.02)
Number of Living Children		
0	1.00	1.00
1	1.05 (0.91, 1.20)	0.97 (0.95, 1.00)
≥ 2	0.95 (0.81, 1.11)	0.97 (0.94, 1.00)
Marital Status		
Married	1.00	1.00
Not married	0.91 (0.80, 1.03)	1.03 (1.00, 1.05)*
Initiation of Prenatal Care in 1st Trimester		
Yes	1.00	1.00
No	0.94 (0.82, 1.08)	0.92 (0.90, 0.95)*
Child		
Birthweight		
< 2,500 grams	1.30 (1.07, 1.57)*	1.17 (1.12, 1.23)*
≥ 2,500 grams	1.00	1.00
Preterm Birth		
< 37 weeks	1.16 (0.96, 1.41)	1.16 (1.11, 1.21)*
≥ 37 weeks	1.00	1.00
Gender		
Female	1.00	1.00
Male	1.07 (0.95, 1.19)	1.08 (1.05, 1.10)*

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)	Children without Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
System		
Source of Prenatal Care		
Health department	1.00	1.00
Other	0.94 (0.83, 1.07)	0.97 (0.95, 1.00)[*]
Receipt of Maternity Care Coordination Services		
Yes	1.18 (1.03, 1.34)[*]	1.05 (1.03, 1.08)[*]
No	1.00	1.00
Receipt of WIC		
Yes	0.87 (0.76, 1.00)	1.04 (1.02, 1.07)[*]
No	1.00	1.00
Birth Hospital Level of Care		
Level III	1.11 (0.98, 1.27)	0.95 (0.92, 0.97)[*]
Community	1.00	1.00
Perinatal Care Region		
Northwestern	1.00	1.00
Southwestern	1.04 (0.86, 1.24)	0.83 (0.80, 0.87)[*]
Northeastern	1.30 (1.09, 1.55)[*]	1.08 (1.05, 1.12)[*]
Southeastern	1.22 (1.00, 1.49)	0.97 (0.94, 1.01)
Eastern	1.12 (0.93, 1.35)	1.01 (0.98, 1.05)
Western	1.25 (1.03, 1.54)[*]	1.18 (1.14, 1.23)[*]
Place of Residence		
Metropolitan	1.00	1.00
Micropolitan	1.00 (0.86, 1.16)	0.96 (0.93, 0.99)[*]
Noncore adjacent	0.97 (0.79, 1.19)	0.92 (0.89, 0.96)[*]
Noncore areas not adjacent	0.98 (0.71, 1.35)	0.91 (0.86, 0.97)[*]

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[*] Statistically significant
[†] Adjusted for all covariates in the model

APPENDIX B. DIRECTION FOR ESTIMATED RELATIONSHIPS BETWEEN SELECTED CHARACTERISTICS AND HEALTH SERVICE USE AND COST AMONG CHILDREN WITH AND WITHOUT OROFACIAL CLEFTS DURING THE FIRST YEAR OF LIFE IN NORTH CAROLINA, 1995-2002

Table 1. Comparison of Direction of Effect from Poisson Results by Service Category among Children with Orofacial Clefts[a]

Characteristics	Medical	Inpatient	Outpatient	Mental Health	Home Health	Total
Maternal						
Age						
≤ 20 years old						
21-24 years old	referent	referent	referent	referent	referent	referent
25-29 years old					++*	
≥ 30 years old						
Education						
< High school				++	++	
High school						
> High school	referent	referent	referent	referent	referent	referent
Race/Ethnicity						
White/non-Hispanic	referent	referent	referent	referent	referent	referent
Black/non-Hispanic						
Hispanic				++	--	
Other[b]			++*	++	++	
Number of Living Children						
0	referent	referent	referent	referent	referent	referent
1					++*	
≥ 2						
Marital Status						
Married	referent	referent	referent	referent	referent	referent
Not married		-*			--	
Initiation of Prenatal Care in 1st Trimester						
Yes	referent	referent	referent	referent	referent	referent
No				++*	--	
Child						
Birthweight						
< 2,500 grams	++*	+*			++*	++*
≥ 2,500 grams	referent	referent	referent	referent	referent	referent
Preterm Birth						
< 37 weeks				++*	--	
≥ 37 weeks	referent	referent	referent	referent	referent	referent

Characteristics	Medical	Inpatient	Outpatient	Mental Health	Home Health	Total
Child (cont.)						
Gender						
Female	referent	referent	referent	referent	referent	referent
Male				++	++*	
System						
Source of Prenatal Care						
Health department	referent	referent	referent	referent	referent	referent
Other		-*				
Receipt of Maternity Care Coordination Services						
Yes	+*				++*	+*
No	referent	referent	referent	referent	referent	referent
Receipt of WIC						
Yes	-*				--*	
No	referent	referent	referent	referent	referent	referent
Birth Hospital Level of Care						
Level III			+*		++*	
Community	referent	referent	referent	referent	referent	referent
Perinatal Care Region						
Northwestern	referent	referent	referent	referent	referent	referent
Southwestern				--	++*	
Northeastern	++*			--*	++*	++*
Southeastern	+*			--	++*	
Eastern				--	++	
Western	++*	+*		--	++	+*
Place of Residence						
Metropolitan	referent	referent	referent	referent	referent	referent
Micropolitan					++	
Noncore adjacent				++		
Noncore areas not adjacent				++	++	

[a] Children were continuously enrolled in Medicaid during first year of life and born in North Carolina, 1995-2002

[b] Other includes Native American, Asian/Pacific Islander and other non-White

+ Count ratio greater than referent and $p < 0.05$

- Count ratio less than referent and $p < 0.05$

++ Count ratio greater than 1.30

-- Count ratio less than 0.70

* Significantly significant

Table 2. Comparison of Direction of Effect from Poisson Results by Service Category for among Children without Orofacial Clefts[a]

Characteristics	Medical	Inpatient	Outpatient	Mental Health	Home Health	Total
Maternal						
Age						
≤ 20 years old				--[*]	--	
21-24 years old	referent	referent	referent	referent	referent	referent
25-29 years old						
≥ 30 years old						
Education						
< High school		+[*]	+[*]	++		
High school			+[*]		--[*]	
> High school	referent	referent	referent	referent	referent	referent
Race/Ethnicity						
White/non-Hispanic	referent	referent	referent	referent	referent	referent
Black/non-Hispanic	-[*]		+[*]			-[*]
Hispanic	-[*]		+[*]		--	-[*]
Other[a]					--	
Number of Living Children						
0	referent	referent	referent	referent	referent	referent
1						
≥ 2						
Marital Status						
Married	referent	referent	referent	referent	referent	referent
Not married				++	++[*]	+[*]
Initiation of Prenatal Care in 1st Trimester						
Yes	referent	referent	referent	referent	referent	referent
No	-[*]	-	-[*]			+[*]
Child						
Birthweight						
< 2,500 grams	+[*]	+[*]	+[*]	++[*]	++[*]	+[*]
≥ 2,500 grams	referent	referent	referent	referent	referent	referent
Preterm Birth						
< 37 weeks	+[*]	+[*]	+[*]	++	++[*]	+[*]
≥ 37 weeks	referent	referent	referent	referent	referent	referent
Gender						
Female	referent	referent	referent	referent	referent	referent
Male	+[*]	+[*]	+[*]	++[*]	++[*]	+[*]

Characteristics	Medical	Inpatient	Outpatient	Mental Health	Home Health	Total
System						
Source of Prenatal Care						
Health department	referent	referent	referent	referent	referent	referent
Other	-*	-*	+*			-*
Receipt of Maternity Care Coordination Services						
Yes	+*			++	++	+*
No	referent	referent	referent	referent	referent	referent
Receipt of WIC						
Yes	+*		+*	--		+*
No	referent	referent	referent	referent	referent	referent
Birth Hospital Level of Care						
Level III	-*		+*		++*	-*
Community	referent	referent	referent	referent	referent	referent
Perinatal Care Region						
Northwestern	referent	referent	referent	referent	referent	referent
Southwestern	-*			--	++	-*
Northeastern	+*			--*	--*	+*
Southeastern		+*		--	++	
Eastern					--*	
Western	+*			++		+*
Place of Residence						
Metropolitan	referent	referent	referent	referent	referent	referent
Micropolitan	-*			++		-*
Noncore adjacent	-*		++*	++		-*
Noncore areas not adjacent	-*		++*	++	++	-*

[a] Children were continuously enrolled in Medicaid during first year of life and born in North Carolina, 1995-2002

[b] Other includes Native American, Asian/Pacific Islander and other non-White

+ Count ratio greater than referent and $p < 0.05$

- Count ratio less than referent and $p < 0.05$

++ Count ratio greater than 1.30

-- Count ratio less than 0.70

* Significantly significant

Table 3. Comparison of Direction of Effect from Binary Logit Results for *any* Outpatient, Mental Health, and Home Health Costs among Children with Orofacial Clefts[a]

Characteristics	Outpatient	Mental Health	Home Health
Maternal			
Age			
≤ 20 years old			
21-24 years old	referent	referent	referent
25-29 years old	++		
≥ 30 years old	++		--
Education			
< High school	++*		
High school			
> High school	referent	referent	referent
Race/Ethnicity			
White/non-Hispanic	referent	referent	referent
Black/non-Hispanic	--		
Hispanic		++	
Other[b]	**	++	
Number of Living Children			
0	referent	referent	referent
1	--*	++*	
≥ 2	--		--
Marital Status			
Married	referent	referent	referent
Not married			
Initiation of Prenatal Care in 1st Trimester			
Yes	referent	referent	referent
No		++	
Child			
Birthweight			
< 2,500 grams	**	++	++*
≥ 2,500 grams	referent	referent	referent
Preterm Birth			
< 37 weeks	**	++*	
≥ 37 weeks	referent	referent	referent
Gender			
Female	referent	referent	referent
Male			
System			
Source of Prenatal Care			
Health department	referent	referent	referent
Other	++	++	

Characteristics	Outpatient	Mental Health	Home Health
System (cont.)			
Receipt of Maternity Care Coordination Services			
Yes	++		++*
No	referent	referent	referent
Receipt of WIC			
Yes	--		--*
No	referent	referent	referent
Birth Hospital Level of Care			
Level III			++*
Community	referent	referent	referent
Perinatal Care Region			
Northwestern	referent	referent	referent
Southwestern			++
Northeastern		--	
Southeastern	++	--	++*
Eastern		--	++
Western	++		++
Place of Residence			
Metropolitan	referent	referent	referent
Micropolitan			
Noncore adjacent	--*	++*	
Noncore areas not adjacent	**	++*	

[a] Children were continuously enrolled in Medicaid during first year of life and born in North Carolina, 1995-2002

[b] Other includes Native American, Asian/Pacific Islander and other non-White

+* Increased odds of cost from referent and $p < 0.05$

−* Decreased odds of cost from referent and $p < 0.05$

++ Odds of cost 1.3 or greater

-- Odds of cost 0.7 or less

** Unable to determine because of insufficient cell sizes

Table 4. Comparison of Direction of Effect from Binary Logit Results for *any* Outpatient, Mental Health, and Home Health Costs among Children without Orofacial Clefts[a]

Characteristics	Outpatient	Mental Health	Home Health
Maternal			
Age			
≤ 20 years old		--	
21-24 years old	referent	referent	referent
25-29 years old	-*		
≥ 30 years old	-*		++
Education			
< High school	++*	++	++
High school			
> High school	referent	referent	referent
Race/Ethnicity			
White/non-Hispanic	referent	referent	referent
Black/non-Hispanic	+*		
Hispanic	+*		--
Other[b]		--	
Number of Living Children			
0	referent	referent	referent
1			
≥ 2	-*		--*
Marital Status			
Married	referent	referent	referent
Not married			
Initiation of Prenatal Care in 1st Trimester			
Yes	referent	referent	referent
No	-*	++	
Child			
Birthweight			
< 2,500 grams		++*	++*
≥ 2,500 grams	referent	referent	referent
Preterm Birth			
< 37 weeks	++*	++*	++*
≥ 37 weeks	referent	referent	referent
Gender			
Female	referent	referent	referent
Male	++*	++	++*
System			
Source of Prenatal Care			
Health department	referent	referent	referent
Other			

Characteristics	Outpatient	Mental Health	Home Health
System (cont.)			
Receipt of Maternity Care Coordination Services			
Yes		++*	
No	referent	referent	referent
Receipt of WIC			
Yes	+*	--*	
No	referent	referent	referent
Birth Hospital Level of Care			
Level III			++*
Community	referent	referent	referent
Perinatal Care Region			
Northwestern	referent	referent	referent
Southwestern		-	
Northeastern		--*	--*
Southeastern		--*	
Eastern		--	-_*
Western		++	
Place of Residence			
Metropolitan	referent	referent	referent
Micropolitan			
Noncore adjacent	++*	++	--
Noncore areas not adjacent		++	

[a] Children were continuously enrolled in Medicaid during first year of life and born in North Carolina, 1995-2002

[b] Other includes Native American, Asian/Pacific Islander and other non-White

+* Increased odds of cost from referent and $p < 0.05$

–* Decreased odds of cost from referent and $p < 0.05$

++ Odds of cost 1.3 or greater

-- Odds of cost 0.7 or less

** Unable to determine because of insufficient cell sizes

Table 5. Comparison of Direction of Effect from Ordinary Least Squares Regression Results by Cost Category among Children with Orofacial Clefts who had Costs[a]

Characteristics	Medical	Inpatient	Outpatient	Mental Health	Home Health	Total
Maternal						
Age						
≤ 20 years old			--		++	
21-24 years old	referent	referent	referent	referent	referent	referent
25-29 years old			-*		++	
≥ 30 years old	-*	--	--	--		
Education						
< High school			++			+*
High school						
> High school	referent	referent	referent	referent	referent	referent
Race/Ethnicity						
White/non-Hispanic	referent	referent	referent	referent	referent	referent
Black/non-Hispanic	-*			++	++	
Hispanic						
Other[b]			++		++	
Number of Living Children						
0	referent	referent	referent	referent	referent	referent
1				--	+*	
≥ 2					+*	
Marital Status						
Married	referent	referent	referent	referent	referent	referent
Not married				++	--	
Initiation of Prenatal Care in 1st Trimester						
Yes	referent	referent	referent	referent	referent	referent
No					--	
Child						
Birthweight						
< 2,500 grams	++	+*		--	+*	+*
≥ 2,500 grams	referent	referent	referent	referent	referent	referent
Preterm Birth						
< 37 weeks	+*	+*		++	-*	+*
≥ 37 weeks	referent	referent	referent	referent	referent	referent
Gender						
Female	referent	referent	referent	referent	referent	referent
Male			+*		+*	

Characteristics	Medical	Inpatient	Outpatient	Mental Health	Home Health	Total
System						
Source of Prenatal Care						
Health department	referent	referent	referent	referent	referent	referent
Other				--	-*	
Receipt of Maternity Care Coordination Services						
Yes	+*	+*			++	
No	referent	referent	referent	referent	referent	referent
Receipt of WIC						
Yes					-*	
No	referent	referent	referent	referent	referent	referent
Birth Hospital Level of Care						
Level III	+*	+*			++	
Community	referent	referent	referent	referent	referent	referent
Perinatal Care Region						
Northwestern	referent	referent	referent	referent	referent	referent
Southwestern				--	+*	
Northeastern				--	++	
Southeastern						
Eastern					++	
Western			-*		++	
Place of Residence						
Metropolitan	referent	referent	referent	referent	referent	referent
Micropolitan						
Noncore adjacent			--		--	-*
Noncore areas not adjacent		++	--		--	

[a] Children were continuously enrolled in Medicaid during first year of life and born in North Carolina, 1995-2002

[b] Other includes Native American, Asian/Pacific Islander and other non-White

+* Cost ratio greater than referent and $p < 0.05$

-* Cost ratio less than referent and $p < 0.05$

++ Cost ratio greater than 1.3

-- Cost ratio less than 0.7

Table 6. Comparison of Direction of Effect from Ordinary Least Squares Regression Results by Cost Category among Children without Orofacial Clefts who had Costs[a]

Characteristics	Medical	Inpatient	Out-patient	Mental Health	Home Health	Total
Maternal						
Age						
≤ 20 years old						
21-24 years old	referent	referent	referent	referent	referent	referent
25-29 years old				--		
≥ 30 years old						
Education						
< High school	+*	+*	+*	++	--	+*
High school				++	--	
> High school	referent	referent	referent	referent	referent	referent
Race/Ethnicity						
White/non-Hispanic	referent	referent	referent	referent	referent	referent
Black/non-Hispanic				--		
Hispanic	-*			--		
Other[b]			+*	--	--	
Number of Living Children						
0	referent	referent	referent	referent	referent	referent
1	-*				--	
≥ 2	-*					
Marital Status						-*
Married	referent	referent	referent	referent	referent	referent
Not married	+*					+*
Initiation of Prenatal Care in 1st Trimester						
Yes	referent	referent	referent	referent	referent	referent
No	-*		-*			-*
Child						
Birthweight						
< 2,500 grams	+*	+*	+*		+*	+*
≥ 2,500 grams	referent	referent	referent	referent	referent	referent
Preterm Birth						
< 37 weeks	+*	+*				+*
≥ 37 weeks	referent	referent	referent	referent	referent	referent
Gender						
Female	referent	referent	referent	referent	referent	referent
Male	+*	+*	+*			+*

Characteristics	Medical	Inpatient	Out-patient	Mental Health	Home Health	Total
System						
Source of Prenatal Care						
Health department	referent	referent	referent	referent	referent	referent
Other			+*	++		
Receipt of Maternity Care Coordination Services						
Yes	+*				++	+*
No	referent	referent	referent	referent	referent	referent
Receipt of WIC						
Yes	+*		+*			+*
No	referent	referent	referent	referent	referent	referent
Birth Hospital Level of Care						
Level III		+*	+*			+*
Community	referent	referent	referent	referent	referent	referent
Perinatal Care Region						
Northwestern	referent	referent	referent	referent	referent	referent
Southwestern	+*					+*
Northeastern		+*	+*	--	++	+*
Southeastern	+*					
Eastern		+*	-*		--	
Western	+*					+*
Place of Residence						
Metropolitan	referent	referent	referent	referent	referent	referent
Micropolitan			-*			
Noncore adjacent						
Noncore areas not adjacent	+			--	++	

[a] Children were continuously enrolled in Medicaid during first year of life and born in North Carolina, 1995-2002

[b] Other includes Native American, Asian/Pacific Islander and other non-White

+* Cost ratio greater than referent and $p < 0.05$

-* Cost ratio less than referent and $p < 0.05$

++ Cost ratio greater than 1.3

-- Cost ratio less than 0.7

APPENDIX C. PREDICTORS OF HEALTH SERVICE USE AND COST, INCLUDING CLEFT TYPE AND PRESENCE OF OTHER BIRTH DEFECTS AMONG CHILDREN WITH OROFACIAL CLEFTS DURING THE FIRST YEAR OF LIFE IN NORTH CAROLINA, 1995-2002

Table 1. Poisson Regression of Selected Characteristics on the Number of Medical Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	0.98 (0.85, 1.12)
21-24 years old	1.00
25-29 years old	0.97 (0.85, 1.10)
≥ 30 years old	0.94 (0.79, 1.10)
Education	
< High school	1.08 (0.93, 1.26)
High school	1.07 (0.93, 1.23)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	1.02 (0.89, 1.16)
Hispanic	0.86 (0.73, 1.02)
Other[b]	1.16 (0.91, 1.47)
Number of Living Children	
0	1.00
1	1.05 (0.94, 1.19)
≥ 2	0.98 (0.85, 1.13)
Marital Status	
Married	1.00
Not married	0.93 (0.83, 1.04)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	0.89 (0.79, 1.01)
Child	
Birthweight	
< 2,500 grams	1.21 (1.03, 1.45)*
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	1.14 (0.96, 1.35)
≥ 37 weeks	1.00

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Gender	
Female	1.00
Male	1.08 (0.97, 1.19)
Cleft Type	
Cleft lip	1.00
Cleft palate	1.19 (1.02, 1.39)[*]
Cleft lip with cleft palate	1.33 (1.15, 1.54)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	1.61 (1.45, 1.79)[*]
System	
Source of Prenatal Care	
Health department	1.00
Other	0.93 (0.83, 1.04)
Receipt of Maternity Care Coordination Services	
Yes	1.17 (1.04, 1.31)[*]
No	1.00
Receipt of WIC	
Yes	0.87 (0.77, 0.98)[*]
No	1.00
Birth Hospital Level of Care	
Level III	1.00 (0.89, 1.12)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	1.04 (0.86, 1.26)
Northeastern	1.35 (1.12, 1.62)[*]
Southeastern	1.25 (1.02, 1.55)
Eastern	1.14 (0.94, 1.38)
Western	1.31 (1.07, 1.62)[*]
Place of Residence	
Metropolitan	1.00
Micropolitan	1.01 (0.88, 1.15)
Noncore adjacent	0.97 (0.82, 1.16)
Noncore areas not adjacent	0.83 (0.62, 1.10)

[a] Children were continuously enrolled in Medicaid during the first year of life

[b] Other includes Native American, Asian/Pacific Islander and other non-White

[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis

[*] Statistically significant

[†] Adjusted for all covariates in the model

Table 2. Poisson Regression of Selected Characteristics on the Number of Inpatient Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	1.02 (0.89, 1.16)
21-24 years old	1.00
25-29 years old	0.96 (0.84, 1.09)
≥ 30 years old	0.87 (0.74, 1.03)
Education	
< High school	1.10 (0.95, 1.27)
High school	1.06 (0.92, 1.21)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	0.94 (0.82, 1.08)
Hispanic	0.83 (0.70, 0.98)
Other[b]	1.26 (1.00, 1.57)*
Number of Living Children	
0	1.00
1	0.98 (0.87, 1.10)
≥ 2	1.04 (0.91, 1.18)
Marital Status	
Married	1.00
Not married	0.87 (0.79, 0.76)*
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	0.93 (0.82, 1.05)
Child	
Birthweight	
< 2,500 grams	1.16 (0.98, 1.37)*
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	1.06 (0.90, 1.26)
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	0.99 (0.89, 1.09)

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	1.28 (1.10, 1.49)[*]
Cleft lip with cleft palate	1.49 (1.29, 1.73)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	1.61 (1.45, 1.78)[*]
System	
Source of Prenatal Care	
Health department	1.00
Other	0.86 (0.77, 0.96)[*]
Receipt of Maternity Care Coordination Services	
Yes	0.99 (0.89, 1.11)
No	1.00
Receipt of WIC	
Yes	0.96 (0.86, 1.08)
No	1.00
Birth Hospital Level of Care	
Level III	1.01 (0.90, 1.13)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	0.94 (0.81, 1.10)
Northeastern	0.93 (0.80, 1.09)
Southeastern	1.03 (0.87, 1.22)
Eastern	1.13 (0.97, 1.32)
Western	1.24 (1.05, 1.47)[*]
Place of Residence	
Metropolitan	1.00
Micropolitan	1.06 (0.94, 1.20)
Noncore adjacent	1.05 (0.89, 1.25)
Noncore areas not adjacent	0.98 (0.76, 1.25)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 3. Poisson Regression of Selected Characteristics on the Number of Outpatient Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	0.87 (0.72, 1.06)
21-24 years old	1.00
25-29 years old	0.91 (0.75, 1.10)
≥ 30 years old	1.01 (0.80, 1.26)
Education	
< High school	1.20 (0.97, 1.50)
High school	1.20 (0.98, 1.47)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	0.94 (0.78, 1.15)
Hispanic	0.96 (0.77, 1.21)
Other[b]	1.56 (1.14, 2.14)[*]
Number of Living Children	
0	1.00
1	0.96 (0.81, 1.14)
≥ 2	0.86 (0.71, 1.06)
Marital Status	
Married	1.00
Not married	0.99 (0.85, 1.16)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	0.86 (0.72, 1.03)
Child	
Birthweight	
< 2,500 grams	1.15 (0.91, 1.47)
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	1.12 (0.88, 1.42)
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	1.10 (0.95, 1.27)

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	1.36 (1.08, 1.71)[*]
Cleft lip with cleft palate	1.72 (1.38, 2.15)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	1.70 (1.46, 1.97)[*]
Source of Prenatal Care	
Health department	1.00
Other	0.95 (0.80, 1.11)
Receipt of Maternity Care Coordination Services	
Yes	1.07 (0.91, 1.25)
No	1.00
Receipt of WIC	
Yes	1.00 (0.84, 1.18)
No	1.00
Birth Hospital Level of Care	
Level III	1.11 (0.94, 1.31)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	0.91 (0.73, 1.15)
Northeastern	1.17 (0.94, 1.44)
Southeastern	1.03 (0.80, 1.31)
Eastern	0.95 (0.75, 1.21)
Western	0.94 (0.72, 1.22)
Place of Residence	
Metropolitan	1.00
Micropolitan	0.86 (0.71, 1.04)
Noncore adjacent	0.88 (0.68, 1.16)
Noncore areas not adjacent	0.75 (0.48, 1.17)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 4. Poisson Regression of Selected Characteristics on the Number of Mental Health Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	1.13 (0.62, 2.07)
21-24 years old	1.00
25-29 years old	1.07 (0.61, 1.88)
≥ 30 years old	0.85 (0.39, 1.87)
Education	
< High school	1.49 (0.73, 3.04)
High school	1.15 (0.58, 2.28)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	0.96 (0.50, 1.84)
Hispanic	1.55 (0.82, 2.93)
Other[b]	1.98 (0.77, 5.05)
Number of Living Children	
0	1.00
1	1.05 (0.61, 1.80)
≥ 2	0.93 (0.50, 1.73)
Marital Status	
Married	1.00
Not married	0.98 (0.60, 1.61)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	1.72 (1.05, 2.81)*
Child	
Birthweight	
< 2,500 grams	1.05 (0.54, 2.03)
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	2.65 (1.40, 5.03)*
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	1.46 (0.91, 2.33)

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	3.23 (1.17, 8.91)[*]
Cleft lip with cleft palate	4.14 (1.54, 11.12)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	2.41 (1.49, 3.87)[*]
System	
Source of Prenatal Care	
Health department	1.00
Other	1.18 (0.71, 1.95)
Receipt of Maternity Care Coordination Services	
Yes	0.83 (0.50, 1.38)
No	1.00
Receipt of WIC	
Yes	1.18 (0.70, 2.01)
No	1.00
Birth Hospital Level of Care	
Level III	0.77 (0.45, 1.32)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	0.62 (0.31, 1.21)
Northeastern	0.40 (0.19, 0.86)[*]
Southeastern	0.44 (0.21, 0.95)[*]
Eastern	0.69 (0.35, 1.39)
Western	0.62 (0.28, 1.38)
Place of Residence	
Metropolitan	1.00
Micropolitan	0.83 (0.45, 1.52)
Noncore adjacent	2.28 (1.16, 4.46)[*]
Noncore areas not adjacent	1.70 (0.60, 4.77)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 5. Poisson Regression of Selected Characteristics on the Number of Home Health Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	1.93 (0.93, 3.97)
21-24 years old	1.00
25-29 years old	1.62 (0.93, 2.81)
≥ 30 years old	0.81 (0.32, 2.05)
Education	
< High school	1.18 (0.56, 2.50)
High school	1.03 (0.52, 2.07)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	1.09 (0.54, 2.19)
Hispanic	0.45 (0.17, 1.18)
Other[b]	1.32 (0.51, 3.45)
Number of Living Children	
0	1.00
1	2.64 (1.52, 4.59)*
≥ 2	1.43 (0.68, 3.00)
Marital Status	
Married	1.00
Not married	0.76 (0.43, 1.35)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	0.45 (0.24, 0.84)*
Child	
Birthweight	
< 2,500 grams	2.24 (1.03, 4.89)*
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	0.56 (0.24, 1.29)
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	2.57 (1.54, 4.28)*

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	14.76 (1.98, 110.27)[*]
Cleft lip with cleft palate	19.47 (2.66, 142.60)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	10.59 (5.49, 20.43)[*]
System	
Source of Prenatal Care	
Health department	1.00
Other	0.86 (0.49, 1.53)
Receipt of Maternity Care Coordination Services	
Yes	1.84 (1.07, 3.16)[*]
No	1.00
Receipt of WIC	
Yes	0.55 (0.32, 0.96)[*]
No	1.00
Birth Hospital Level of Care	
Level III	1.42 (0.82, 2.45)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	4.83 (2.07, 11.23)[*]
Northeastern	3.95 (1.65, 9.45)[*]
Southeastern	2.29 (0.85, 6.17)
Eastern	2.25 (0.86, 5.89)
Western	1.52 (0.51, 4.49)
Place of Residence	
Metropolitan	1.00
Micropolitan	2.03 (1.17, 3.52)[*]
Noncore adjacent	1.64 (0.72, 3.76)
Noncore areas not adjacent	1.05 (0.24, 4.53)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 6. Poisson Regression of Selected Characteristics on the Number of Total Paid Claims among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Count Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	0.98 (0.86, 1.12)
21-24 years old	1.00
25-29 years old	0.98 (0.87, 1.11)
≥ 30 years old	0.95 (0.81, 1.11)
Education	
< High school	1.10 (0.96, 1.27)
High school	1.09 (0.95, 1.24)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	1.01 (0.89, 1.15)
Hispanic	0.88 (0.75, 1.03)
Other[b]	1.21 (0.97, 1.51)
Number of Living Children	
0	1.00
1	1.05 (0.94, 1.18)
≥ 2	0.96 (0.84, 1.10)
Marital Status	
Married	1.00
Not married	0.93 (0.84, 1.04)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	0.90 (0.80, 1.01)
Child	
Birthweight	
< 2,500 grams	1.20 (1.02, 1.41)*
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	1.15 (0.98, 1.35)
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	1.09 (0.99, 1.20)

Characteristics	Children with Orofacial Clefts Count Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	1.22 (1.06, 1.41)[*]
Cleft lip with cleft palate	1.38 (1.20, 1.59)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	1.64 (1.49, 1.81)[*]
System	
Source of Prenatal Care	
Health department	1.00
Other	0.94 (0.84, 1.05)
Receipt of Maternity Care Coordination Services	
Yes	1.14 (1.02, 1.27)[*]
No	1.00
Receipt of WIC	
Yes	0.89 (0.79, 0.99)[*]
No	1.00
Birth Hospital Level of Care	
Level III	1.01 (0.90, 1.12)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	1.02 (0.87, 1.18)
Northeastern	1.25 (1.08, 1.44)[*]
Southeastern	1.13 (0.95, 1.33)
Eastern	1.13 (0.97, 1.33)
Western	1.25 (1.05, 1.48)[*]
Place of Residence	
Metropolitan	1.00
Micropolitan	1.01 (0.89, 1.14)
Noncore adjacent	0.99 (0.84, 1.18)
Noncore areas not adjacent	0.85 (0.65, 1.11)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 7. Binary Logit Regression of Selected Characteristics on *Any* Outpatient, Mental Health, and Home Health Costs among Children with and without Orofacial Clefts during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics (n=563)	**Outpatient (n=563)**	**Mental Health (n=563)**	**Home Health**
	Odds Ratio[†] (95% CI)	Odds Ratio[†] (95% CI)	Odds Ratio[†] (95% CI)
Maternal			
Age			
≤ 20 years old	0.62 (0.22, 1.77)	1.11 (0.57, 2.17)	1.15 (0.58, 2.26)
21-24 years old	1.00	1.00	1.00
25-29 years old	2.25 (0.80, 6.33)	1.00 (0.53, 1.89)	1.08 (0.56, 2.10)
≥ 30 years old	2.02 (0.61, 6.72)	1.58 (0.74, 3.37)	0.73 (0.30, 1.76)
Education			
< High school	4.14 (1.30, 13.16)*	1.21 (0.58, 2.51)	1.12 (0.53, 2.41)
High school	0.99 (0.38, 2.59)	1.22 (0.61, 2.44)	1.05 (0.51, 2.14)
> High school	1.00	1.00	1.00
Race			
White/non-Hispanic	1.00	1.00	1.00
Black/non-Hispanic	0.63 (0.24, 1.66)	0.74 (0.37, 1.46)	0.78 (0.39, 1.56)
Hispanic	0.89 (0.24, 3.25)	1.46 (0.70, 3.08)	0.69 (0.30, 1.59)
Other[b]	**	2.97 (0.91, 9.70)	0.88 (0.26, 2.97)
Number of Living Children			
0	1.00	1.00	1.00
1	0.30 (0.12, 0.76)*	2.00 (1.13, 3.56)*	1.18 (0.66, 2.12)
≥ 2	0.40 (0.13, 1.27)	0.97 (0.48, 1.93)	0.61 (0.29, 1.27)
Marital Status			
Married	1.00	1.00	1.00
Not married	0.84 (0.36, 1.97)	1.08 (0.64, 1.83)	0.99 (0.57, 1.73)
Initiation of Prenatal Care in First Trimester			
Yes	1.00	1.00	1.00
No	1.25 (0.46, 3.39)	1.61 (0.92, 2.83)	0.77 (0.42, 1.44)
Child			
Birthweight			
< 2,500 grams	**	1.23 (0.55, 2.74)	2.33 (1.04, 5.25)*
≥ 2,500 grams	1.00	1.00	1.00
Preterm Birth			
< 37 weeks	**	2.28 (1.04, 5.02)*	0.95 (0.41, 2.21)
≥ 37 weeks	1.00	1.00	1.00
Gender			
Female	1.00	1.00	1.00
Male	0.90 (0.43, 1.88)	1.08 (0.66, 1.76)	1.38 (0.82, 2.32)
Cleft Type			
Cleft lip	1.00	1.00	1.00
Cleft palate	0.90 (0.38, 2.14)	2.35 (0.94, 5.87)	3.59 (1.38, 9.31)*
Cleft lip with cleft palate	3.09 (1.15, 8.32)*	7.11 (2.96, 17.07)*	4.23 (1.67, 10.73)*
Presence of Other Anomalies[c]			
Isolated anomaly	1.00	1.00	1.00
Multiple anomalies	1.50 (0.64, 3.50)	3.89 (2.32, 6.54)*	3.92 (2.33, 6.58)*

Characteristics (n=563)	**Outpatient (n=563)**	**Mental Health (n=563)**	**Home Health**
	Odds Ratio[†] (95% CI)	Odds Ratio[†] (95% CI)	Odds Ratio[†] (95% CI)
System			
Source of Prenatal Care			
Health department	1.00	1.00	1.00
Other	1.28 (0.54, 3.03)	1.61 (0.92, 2.79)	1.04 (0.58, 1.84)
Receipt of Maternity Care Coordination Services			
Yes	1.63 (0.66, 4.03)	1.23 (0.72, 2.11)	1.75 (0.98, 3.12)
No	1.00	1.00	1.00
Receipt of WIC			
Yes	0.41 (0.15, 1.13)	1.33 (0.76, 2.33)	0.52 (0.29, 0.95)[*]
No	1.00	1.00	1.00
Birth Hospital Level of Care			
Level III	0.84 (0.34, 2.10)	0.79 (0.45, 1.39)	1.39 (0.78, 2.49)
Community	1.00	1.00	1.00
Perinatal Care Region			
Northwestern	1.00	1.00	1.00
Southwestern	0.63 (0.21, 1.88)	0.83 (0.41, 1.66)	1.93 (0.88, 4.23)
Northeastern	1.21 (0.36, 4.05)	0.39 (0.18, 0.87)[*]	1.20 (0.51, 2.29)
Southeastern	2.21 (0.49, 0.97)	0.32 (0.13, 0.77)[*]	2.97 (1.26, 6.99)[*]
Eastern	1.36 (0.44, 4.21)	0.55 (0.25, 1.22)	1.82 (0.78, 4.28)
Western	1.06 (0.24, 4.60)	0.71 (0.30, 1.69)	1.96 (0.79, 4.85)
Place of Residence			
Metropolitan	1.00	1.00	1.00
Micropolitan	0.89 (0.34, 2.32)	0.97 (0.52, 1.81)	1.25 (0.65, 2.40)
Noncore adjacent	0.42 (0.12, 1.49)	3.72 (1.66, 8.34)[*]	1.34 (0.56, 3.21)
Noncore areas not adjacent	**	2.61 (0.82, 8.33)	0.79 (0.18, 3.38)

[a] Children were continuously enrolled in Medicaid during the first year of life

[b] Other includes Native American, Asian/Pacific Islander and other non-White

[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis

[*] $P < 0.05$

** Unable to determine estimate due to insufficient cell sizes

[†] Adjusted for all covariates in the model

Table 8. Ordinary Least Squares Regression of Selected Characteristics on Medical Costs among Children with and without Orofacial Clefts with Medical Costs during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Cost Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	0.92 (0.75, 1.13)
21-24 years old	1.00
25-29 years old	0.85 (0.70, 1.04)
≥ 30 years old	0.77 (0.61, 0.97)*
Education	
< High school	1.18 (0.94, 1.47)
High school	1.02 (0.83, 1.26)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	0.78 (0.64, 0.95)*
Hispanic	0.85 (0.67, 1.08)
Other[b]	1.26 (0.86, 1.85)
Number of Living Children	
0	1.00
1	1.01 (0.85, 1.21)
≥ 2	1.05 (0.86, 1.28)
Marital Status	
Married	1.00
Not married	0.94 (0.80, 1.11)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	1.00 (0.84, 1.19)
Child	
Birthweight	
< 2,500 grams	1.21 (0.92, 1.58)
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	1.39 (1.06, 1.82)*
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	1.13 (0.98, 1.31)

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	1.10 (0.90, 1.36)
Cleft lip with cleft palate	1.64 (1.35, 2.00)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	1.96 (1.67, 2.30)[*]
System	
Source of Prenatal Care	
Health department	1.00
Other	0.88 (0.74, 1.05)
Receipt of Maternity Care Coordination Services	
Yes	1.19 (1.00, 1.41)
No	1.00
Receipt of WIC	
Yes	0.90 (0.76, 1.07)
No	1.00
Birth Hospital Level of Care	
Level III	1.11 (0.93, 1.32)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	0.97 (0.77, 1.22)
Northeastern	0.98 (0.78, 1.24)
Southeastern	1.12 (0.86, 1.45)
Eastern	1.06 (0.84, 1.35)
Western	1.10 (0.84, 1.45)
Place of Residence	
Metropolitan	1.00
Micropolitan	0.98 (0.81, 1.19)
Noncore adjacent	0.84 (0.64, 1.10)
Noncore areas not adjacent	1.03 (0.69, 1.55)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 9. Ordinary Least Squares Regression of Selected Characteristics on Inpatient Costs among Children with and without Orofacial Clefts with Inpatient Costs during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=550)** Cost Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	1.13 (0.85, 1.49)
21-24 years old	1.00
25-29 years old	1.03 (0.78, 1.36)
≥ 30 years old	0.74 (0.53, 1.03)
Education	
< High school	1.11 (0.82, 1.51)
High school	1.06 (0.79, 1.41)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	0.80 (0.60, 1.05)
Hispanic	0.80 (0.57, 1.12)
Other[b]	1.17 (0.69, 1.98)
Number of Living Children	
0	1.00
1	1.16 (0.90, 1.48)
≥ 2	1.26 (0.95, 1.67)
Marital Status	
Married	1.00
Not married	0.88 (0.70, 1.10)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	0.92 (0.72, 1.18)
Child	
Birthweight	
< 2,500 grams	1.64 (1.13, 2.40)*
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	1.69 (1.16, 2.47)*
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	1.00 (0.82, 1.23)

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	1.61 (1.21, 2.15)[*]
Cleft lip with cleft palate	1.96 (1.49, 2.58)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	2.91 (2.33, 3.64)[*]
System	
Source of Prenatal Care	
Health department	1.00
Other	0.88 (0.70, 1.12)
Receipt of Maternity Care Coordination Services	
Yes	1.24 (0.98, 1.58)
No	1.00
Receipt of WIC	
Yes	0.93 (0.73, 1.19)
No	1.00
Birth Hospital Level of Care	
Level III	1.32 (1.03, 1.67)[*]
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	1.0 (0.73, 1.38)
Northeastern	0.97 (0.70, 1.35)
Southeastern	1.05 (0.73, 1.52)
Eastern	1.18 (0.85, 1.63)
Western	1.14 (0.78, 1.66)
Place of Residence	
Metropolitan	1.00
Micropolitan	1.01 (0.78, 1.32)
Noncore adjacent	1.02 (0.70, 1.50)
Noncore areas not adjacent	1.02 (0.58, 1.78)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 10. Ordinary Least Squares Regression of Selected Characteristics on Outpatient Costs among Children with and without Orofacial Clefts with Outpatient Costs during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=513)** Cost Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	0.61 (0.41, 0.91)[*]
21-24 years old	1.00
25-29 years old	0.51 (0.35, 0.76)[*]
≥ 30 years old	0.79 (0.49, 1.25)
Education	
< High school	1.57 (1.01, 2.42)[*]
High school	1.26 (0.83, 1.91)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	1.00 (0.67, 1.49)
Hispanic	1.02 (0.64, 1.63)
Other[b]	1.70 (0.82, 3.49)
Number of Living Children	
0	1.00
1	0.93 (0.65, 1.33)
≥ 2	0.84 (0.56, 1.25)
Marital Status	
Married	1.00
Not married	1.06 (0.77, 1.45)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	1.06 (0.75, 1.51)
Child	
Birthweight	
< 2,500 grams	1.10 (0.66, 1.83)
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	1.16 (0.67, 1.86)
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	1.22 (0.91 1.64)

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	0.48 (0.32, 0.74)[*]
Cleft lip with cleft palate	1.80 (1.21, 2.66)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	1.27 (0.92, 1.74)
System	
Source of Prenatal Care	
Health department	1.00
Other	0.84 (0.60, 1.18)
Receipt of Maternity Care Coordination Services	
Yes	0.88 (0.63, 1.22)
No	1.00
Receipt of WIC	
Yes	0.89 (0.63, 1.25)
No	1.00
Birth Hospital Level of Care	
Level III	0.70 (0.50, .99)[*]
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	0.73 (0.46, 1.14)
Northeastern	1.13 (0.71, 1.79)
Southeastern	1.01 (0.61, 1.68)
Eastern	0.50 (0.31, 0.79)[*]
Western	0.26 (0.15, 0.44)[*]
Place of Residence	
Metropolitan	1.00
Micropolitan	0.79 (0.54, 1.15)
Noncore adjacent	0.51 (0.29, 0.87)[*]
Noncore areas not adjacent	0.51 (0.23, 1.11)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 11. Ordinary Least Squares Regression of Selected Characteristics on Mental Health Costs among Children with and without Orofacial Clefts with Mental Health Costs during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=121)** Cost Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	0.85 (0.40, 1.81)
21-24 years old	1.00
25-29 years old	0.77 (0.38, 1.57)
≥ 30 years old	0.53 (0.24, 1.19)
Education	
< High school	1.19 (0.53, 2.68)
High school	0.75 (0.33, 1.72)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	1.34 (0.62, 2.88)
Hispanic	0.79 (0.33, 1.86)
Other[b]	0.89 (0.28, 2.83)
Number of Living Children	
0	1.00
1	0.66 (0.35, 1.27)
≥ 2	1.08 (0.49, 2.36)
Marital Status	
Married	1.00
Not married	1.48 (0.81, 2.70)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	1.27 (0.68, 2.37)
Child	
Birthweight	
< 2,500 grams	0.40 (0.15, 1.07)
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	2.21 (0.86, 5.72)
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	1.24 (0.71, 2.16)

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	1.38 (0.44, 4.39)
Cleft lip with cleft palate	1.26 (0.41, 3.90)
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	1.73 (0.96, 3.13)
System	
Source of Prenatal Care	
Health department	1.00
Other	0.60 (0.33, 1.11)
Receipt of Maternity Care Coordination Services	
Yes	0.84 (0.47, 1.49)
No	1.00
Receipt of WIC	
Yes	0.79 (0.42, 1.49)
No	1.00
Birth Hospital Level of Care	
Level III	0.98 (0.53, 1.83)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	0.43 (0.20, 0.93)[*]
Northeastern	0.74 (0.29, 1.86)
Southeastern	0.63 (0.24, 1.68)
Eastern	0.91 (0.35, 2.33)
Western	0.69 (0.26, 1.81)
Place of Residence	
Metropolitan	1.00
Micropolitan	0.98 (0.48, 2.02)
Noncore adjacent	1.27 (0.53, 3.05)
Noncore areas not adjacent	0.79 (0.23, 2.70)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 12. Ordinary Least Squares Regression of Selected Characteristics on Home Health Costs among Children with and without Orofacial Clefts with Home Health Costs during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=101)** Cost Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	2.39 (0.75, 7.61)
21-24 years old	1.00
25-29 years old	1.57 (0.64, 3.91)
≥ 30 years old	0.69 (0.18, 2.63)
Education	
< High school	0.99 (0.31, 3.18)
High school	1.22 (0.43, 3.45)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	1.80 (0.64, 5.02)
Hispanic	0.82 (0.22, 3.01)
Other[b]	2.91 (0.56, 15.26)
Number of Living Children	
0	1.00
1	3.70 (1.42, 9.59)[*]
≥ 2	8.92 (2.36, 33.68)[*]
Marital Status	
Married	1.00
Not married	0.53 (0.22, 1.25)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	0.49 (0.20, 1.18)
Child	
Birthweight	
< 2,500 grams	4.86 (1.60, 14.76)[*]
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	0.21 (0.06, 0.72)[*]
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	3.96 (1.68, 9.39)[*]

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	1.88 (0.37, 9.41)
Cleft lip with cleft palate	1.96 (0.38, 10.10)
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	2.38 (1.06, 5.34)
System	
Source of Prenatal Care	
Health department	1.00
Other	0.37 (0.15, 0.91)[*]
Receipt of Maternity Care Coordination Services	
Yes	1.29 (0.54, 3.05)
No	1.00
Receipt of WIC	
Yes	0.33 (0.13, 0.79)[*]
No	1.00
Birth Hospital Level of Care	
Level III	1.71 (0.68, 4.33)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	4.35 (1.32, 14.35)[*]
Northeastern	2.66 (0.73, 9.65)
Southeastern	1.19 (0.37, 3.81)
Eastern	1.88 (0.51, 6.85)
Western	1.95 (0.56, 6.70)
Place of Residence	
Metropolitan	1.00
Micropolitan	1.32 (0.51, 3.43)
Noncore adjacent	0.44 (0.13, 1.49)
Noncore areas not adjacent	0.23 (0.02, 2.15)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

Table 13. Ordinary Least Squares Regression of Selected Characteristics on Total Costs among Children with and without Orofacial Clefts with Total Costs during the First Year of Life in North Carolina, 1995-2002[a]

Characteristics	**Children with Orofacial Clefts (n=563)** Cost Ratios[†] (95% Confidence Intervals)
Maternal	
Age	
≤ 20 years old	0.91 (0.72, 1.15)
21-24 years old	1.00
25-29 years old	0.87 (0.69, 1.09)
≥ 30 years old	0.80 (0.61, 1.05)
Education	
< High school	1.40 (1.09, 1.80)*
High school	1.16 (0.92, 1.48)
> High school	1.00
Race/Ethnicity	
White/non-Hispanic	1.00
Black/non-Hispanic	0.83 (0.66, 1.05)
Hispanic	0.82 (0.62, 1.08)
Other[b]	1.34 (0.87, 2.09)
Number of Living Children	
0	1.00
1	0.98 (0.80, 1.21)
≥ 2	0.99 (0.78, 1.25)
Marital Status	
Married	1.00
Not married	0.92 (0.76, 1.11)
Initiation of Prenatal Care in 1st Trimester	
Yes	1.00
No	1.02 (0.83, 1.26)
Child	
Birthweight	
< 2,500 grams	1.42 (1.04, 1.04)*
≥ 2,500 grams	1.00
Preterm Birth	
< 37 weeks	1.44 (1.05, 1.96)*
≥ 37 weeks	1.00
Gender	
Female	1.00
Male	1.11 (0.93, 1.31)

Characteristics	Children with Orofacial Clefts Cost Ratios[†] (95% Confidence Intervals)
Child (cont.)	
Cleft Type	
Cleft lip	1.00
Cleft palate	1.11 (0.87, 1.41)
Cleft lip with cleft palate	1.26 (0.41, 3.90)[*]
Presence of Other Anomalies[c]	
Isolated anomaly	1.00
Multiple anomalies	2.22 (1.85, 2.67)[*]
System	
Source of Prenatal Care	
Health department	1.00
Other	0.90 (0.74, 1.09)
Receipt of Maternity Care Coordination Services	
Yes	1.18 (0.97, 1.44)
No	1.00
Receipt of WIC	
Yes	0.90 (0.73, 1.10)
No	1.00
Birth Hospital Level of Care	
Level III	1.04 (0.86, 1.27)
Community	1.00
Perinatal Care Region	
Northwestern	1.00
Southwestern	0.99 (0.76, 1.28)
Northeastern	1.00 (0.76, 1.30)
Southeastern	1.10 (0.81, 1.48)
Eastern	1.03 (0.78, 1.35)
Western	0.93 (0.68, 1.27)
Place of Residence	
Metropolitan	1.00
Micropolitan	1.01 (0.81, 1.26)
Noncore adjacent	0.70 (0.51, 0.96)[*]
Noncore areas not adjacent	0.89 (0.56, 1.43)

[a] Children were continuously enrolled in Medicaid during the first year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly=orofacial cleft diagnosis only; multiple anomalies=orofacial cleft and other birth defect diagnosis
[*] Statistically significant
[†] Adjusted for all covariates in the model

APPENDIX D: DEMOGRAPHICS OF CHILDREN WITH AND WITHOUT OROFACIAL CLEFTS DURING THE SECOND THROUGH FIFTH YEAR OF LIFE IN NORTH CAROLINA, 1995-2002

Table 1. Selected Characteristics of Children with and without Orofacial Clefts (OFC) during the 2nd Year of Life in North Carolina, 1995-2002[a]

Characteristic	Children with OFC N = 406 (%)	Children without OFC N = 3,692 (%)	P-value
Maternal			
Age			
≤ 20 years old	132 (32.5)	1,316 (36.2)	0.02*
21-24 years old	111 (27.3)	1,091 (30.0)	
25-29 years old	106 (26.1)	706 (19.4)	
≥ 30 years old	57 (14.0)	519 (14.3)	
Education			
< High school	192 (47.3)	1,618 (44.6)	0.57
High school	150 (37.0)	1,423 (39.2)	
> High school	64 (15.8)	591 (16.3)	
Race			
White/non-Hispanic	236 (58.1)	1,478 (46.4)	<0.00*
Black/non-Hispanic	103 (25.4)	1,568 (36.6)	
Hispanic	48 (11.8)	457 (13.3)	
Other[b]	19 (4.7)	129 (3.7)	
Number of Living Children			
0	171 (42.1)	1,585 (43.7)	0.64
1	119 (29.3)	1,088 (30.0)	
≥ 2	116 (28.6)	958 (26.4)	
Marital Status			
Married	170 (41.9)	1,235 (34.0)	0.00*
Not married	236 (58.1)	2,397 (66.0)	
Initiation of Prenatal Care in First Trimester			
Yes	303 (74.6)	2,662 (73.3)	0.79
No	101(24.9)	945 (26.0)	
Child			
Birthweight			
< 2,500 grams	60 (14.8)	340 (9.4)	<0.00*
≥ 2,500 grams	346 (85.2)	3,292 (90.6)	
Preterm Birth			
< 37 weeks	64 (15.8)	427 (11.8)	0.02*
≥ 37 weeks	342 (84.2)	3,205 (88.2)	
Gender			
Female	178 (43.8)	1,780 (49.0)	0.05
Male	228 (56.2)	1,852 (51.0)	

Characteristic	Children with OFC N = 406 (%)	Children without OFC N = 3,632 (%)	P-value
Child (cont.)			
Cleft Type			
Cleft lip	74 (18.2)	n/a	n/a
Cleft palate	150 (37.0)	n/a	
Cleft lip with cleft palate	182 (44.8)	n/a	
Presence of Other Anomalies[c]			
Isolated	260 (64.0)	n/a	n/a
Multiple	146 (36.0)	n/a	
System			
Source of Prenatal Care			
Health department	235 (57.9)	2,051 (56.5)	0.59
Other	171 (42.1)	1,581 (43.5)	
Receipt of Maternity Care Coordination Services			
Yes	194 (47.8)	1,788 (49.2)	0.58
No	212 (52.2)	1,844 (50.8)	
Receipt of WIC			
Yes	286 (70.4)	2,763 (76.1)	0.01*
No	120 (29.6)	869 (23.9)	
Birth Hospital Level of Care			
Level III	180 (44.3)	1,376 (37.9)	0.01*
Community	226 (55.7)	2,256 (62.1)	
Perinatal Care Region			
Northwestern	97 (23.9)	846 (23.3)	0.02*
Southwestern	73 (18.0)	507 (14.0)	
Northeastern	63 (15.5)	589 (16.2)	
Southeastern	56 (13.8)	671 (18.5)	
Eastern	72 (17.7)	722 (19.9)	
Western	45 (11.1)	297 (8.2)	
Place of Residence			
Metropolitan	258 (63.6)	2,225 (61.3)	0.82
Micropolitan	92 (23.7)	1,374 (22.7)	
Noncore areas adjacent to metro area or small town	38 (9.4)	349 (9.6)	
Noncore areas not adjacent to metro area or small town	18 (4.4)	163 (4.5)	

[a] Children were continuously enrolled in Medicaid during the second year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly = cleft lip with or without cleft palate diagnosis only diagnosis; multiple anomalies = cleft lip with or without cleft palate and other birth defect diagnosis
* Statistically significant ($p < 0.05$) by chi-square test

Table 2. Selected Characteristics of Children with and without Orofacial Clefts (OFC) during the 3rd Year of Life in North Carolina, 1995-2002[a]

Characteristic	Children with OFC N = 300 (%)	Children without OFC N = 2,708 (%)	P-value
Maternal			
Age			
≤ 20 years old	99 (33.0)	1,040 (38.4)	0.03*
21-24 years old	87 (29.0)	766 (28.3)	
25-29 years old	77 (25.7)	517 (19.1)	
≥ 30 years old	37 (12.3)	385 (14.2)	
Education			
< High school	148 (49.3)	1,231 (45.5)	0.44
High school	109 (36.3)	1,069 (39.5)	
> High school	43 (14.3)	408 (15.1)	
Race			
White/non-Hispanic	169 (56.3)	1,064 (39.3)	<0.00*
Black/non-Hispanic	82 (27.3)	1,244 (45.9)	
Hispanic	32 (10.7)	298 (11.0)	
Other[b]	17 (4.7)	102 (3.8)	
Number of Living Children			
0	127 (42.3)	1,139 (42.1)	0.99
1	91 (30.3)	831 (30.7)	
≥ 2	82 (27.3)	738 (27.3)	
Marital Status			
Married	120 (40.0)	884 (32.6)	0.01*
Not married	180 (60.0)	1,824 (67.4)	
Initiation of Prenatal Care in First Trimester			
Yes	226 (75.3)	1,961 (72.4)	0.48
No	73 (24.3)	729 (26.9)	
Child			
Birthweight			
< 2,500 grams	47 (15.7)	283 (10.5)	0.01*
≥ 2,500 grams	253 (84.3)	2,425 (89.6)	
Preterm Birth			
< 37 weeks	48 (16.0)	354 (13.1)	0.16
≥ 37 weeks	252 (84.0)	2,354 (86.9)	
Gender			
Female	135 (45.0)	1,336 (49.3)	0.15
Male	165 (55.0)	1,372 (50.7)	

Characteristic	Children with OFC N = 300 (%)	Children without OFC N = 2,708 (%)	P-value
Child (cont.)			
Cleft Type			
Cleft lip	44 (14.7)	n/a	n/a
Cleft palate	119 (39.7)	n/a	
Cleft lip with cleft palate	137 (45.7)	n/a	
Presence of Other Anomalies[c]			
Isolated	181 (60.3)	n/a	n/a
Multiple	119 (39.7)	n/a	
System			
Source of Prenatal Care			
Health department	176 (58.7)	1,515 (56.0)	0.37
Other	124 (41.3)	1,193 (44.1)	
Receipt of Maternity Care Coordination Services			
Yes	147 (49.0)	1,365 (50.4)	0.64
No	153 (51.0)	1,343 (49.6)	
Receipt of WIC			
Yes	208 (69.3)	2,070 (76.4)	0.01*
No	92 (30.7)	638 (23.6)	
Birth Hospital Level of Care			
Level III	139 (46.3)	1,024 (37.8)	0.00*
Community	161 (53.7)	1,684 (62.2)	
Perinatal Care Region			
Northwestern	78 (26.0)	624 (23.0)	0.05
Southwestern	42 (14.0)	370 (13.7)	
Northeastern	44 (14.7)	413 (15.3)	
Southeastern	46 (15.3)	523 (19.3)	
Eastern	51 (17.0)	549 (20.3)	
Western	39 (13.0)	229 (8.5)	
Place of Residence			
Metropolitan	186 (62.0)	1,597 (59.0)	0.70
Micropolitan	75 (25.0)	706 (26.1)	
Noncore areas adjacent to metro area or small town	27 (9.0)	266 (9.8)	
Noncore areas not adjacent to metro area or small town	12 (4.0)	139 (5.1)	

[a] Children were continuously enrolled in Medicaid during the third year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly = cleft lip with or without cleft palate diagnosis only diagnosis; multiple anomalies = cleft lip with or without cleft palate and other birth defect diagnosis
* Statistically significant ($p < 0.05$) by chi-square test

Table 3. Selected Characteristics of Children with and without Orofacial Clefts (OFC) during the 4th Year of Life in North Carolina, 1995-2002[a]

Characteristic	Children with OFC N = 246 (%)	Children without OFC N = 2,135 (%)	P-value
Maternal			
Age			
≤ 20 years old	89 (36.2)	841 (39.4)	0.04*
21-24 years old	60 (24.4)	605 (28.3)	
25-29 years old	64 (26.0)	395 (18.5)	
≥ 30 years old	33 (13.4)	294 (13.8)	
Education			
< High school	128 (52.0)	992 (46.7)	0.25
High school	88 (35.8)	840 (39.3)	
> High school	30 (12.2)	303 (14.2)	
Race			
White/non-Hispanic	142 (57.7)	860 (40.3)	<0.00*
Black/non-Hispanic	67 (27.2)	992 (46.5)	
Hispanic	23 (9.4)	213 (10.0)	
Other[b]	14 (5.7)	70 (3.3)	
Number of Living Children			
0	103 (41.9)	901 (42.2)	0.94
1	72 (29.3)	639 (29.9)	
≥ 2	71 (28.9)	594 (27.8)	
Marital Status			
Married	101 (41.1)	710 (33.3)	0.01*
Not married	145 (58.9)	1,425 (66.7)	
Initiation of Prenatal Care in First Trimester			
Yes	184 (74.8)	1,539 (72.1)	0.67
No	61 (24.8)	586 (27.5)	
Child			
Birthweight			
< 2,500 grams	38 (15.5)	229 (10.7)	0.03*
≥ 2,500 grams	208 (84.6)	1,906 (89.3)	
Preterm Birth			
< 37 weeks	40 (16.3)	275 (12.9)	0.14
≥ 37 weeks	206 (83.7)	1,860 (87.1)	
Gender			
Female	111 (45.1)	1,057 (49.5)	0.19
Male	135 (54.9)	1,078 (50.5)	
Cleft Type			
Cleft lip	36 (14.6)	n/a	n/a
Cleft palate	94 (38.2)	n/a	
Cleft lip with cleft palate	116 (47.2)	n/a	

Characteristic	Children with OFC N = 246 (%)	Children without OFC N = 2,135 (%)	P-value
Child (cont.)			
Presence of Other Anomalies[c]			
Isolated	157 (63.8)	n/a	n/a
Multiple	89 (36.2)	n/a	
System			
Source of Prenatal Care			
Health department	139 (56.5)	1,173 (54.9)	0.64
Other	107 (43.5)	962 (45.1)	
Receipt of Maternity Care Coordination Services			
Yes	121 (49.2)	1,107 (51.9)	0.43
No	125 (50.8)	1,028 (48.2)	
Receipt of WIC			
Yes	174 (70.7)	1,668 (78.1)	0.01*
No	72 (29.3)	467 (21.9)	
Birth Hospital Level of Care			
Level III	114 (46.3)	797 (37.3)	0.01*
Community	132 (53.7)	1,338 (62.7)	
Perinatal Care Region			
Northwestern	58 (23.6)	495 (23.2)	0.15
Southwestern	38 (15.5)	290 (13.6)	
Northeastern	38 (15.5)	325 (15.2)	
Southeastern	38 (15.5)	401 (18.8)	
Eastern	43 (17.5)	449 (21.0)	
Western	31 (12.6)	175 (8.2)	
Place of Residence			
Metropolitan	150 (61.0)	1,264 (59.2)	0.55
Micropolitan	63 (25.6)	563 (26.4)	
Noncore areas adjacent to metro area or small town	25 (10.2)	196 (9.2)	
Noncore areas not adjacent to metro area or small town	8 (3.3)	112 (5.3)	

[a] Children were continuously enrolled in Medicaid during the fourth year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly = cleft lip with or without cleft palate diagnosis only diagnosis; multiple anomalies = cleft lip with or without cleft palate and other birth defect diagnosis
* Statistically significant ($p < 0.05$) by chi-square test

Table 4. Selected Characteristics of Children with and without Orofacial Clefts (OFC) during the 5th Year of Life in North Carolina, 1995-2002[a]

Characteristic	Children with OFC N = 184 (%)	Children without OFC N = 1,704 (%)	P-value
Maternal			
Age			
≤ 20 years old	61 (33.2)	678 (39.8)	0.01*
21-24 years old	44 (23.9)	468 (27.5)	
25-29 years old	54 (29.4)	321 (18.8)	
≥ 30 years old	25 (13.6)	237 (13.9)	
Education			
< High school	92 (50.0)	812 (47.7)	0.71
High school	66 (35.9)	664 (39.0)	
> High school	26 (14.1)	228 (13.4)	
Race			
White/non-Hispanic	114 (62.0)	700 (41.1)	<0.00*
Black/non-Hispanic	48 (26.1)	786 (46.1)	
Hispanic	14 (7.6))	156 (9.2)	
Other[b]	8 (4.4)	62 (3.6)	
Number of Living Children			
0	77 (41.9)	729 (42.8)	0.84
1	54 (29.4)	518 (30.4)	
≥ 2	53 (28.8)	456 (26.8)	
Marital Status			
Married	82 (44.6)	566 (33.2)	0.00*
Not married	102 (55.4)	1,138 (66.8)	
Initiation of Prenatal Care in First Trimester			
Yes	139 (75.5)	1,258 (73.8)	0.84
No	44 (23.9)	439 (25.8)	
Child			
Birthweight			
< 2,500 grams	22 (12.0)	177 (10.4)	0.51
≥ 2,500 grams	162 (88.0)	1,527 (89.6)	
Preterm Birth			
< 37 weeks	24 (13.0)	200 (11.7)	0.60
≥ 37 weeks	160 (87.0)	1,504 (88.3)	
Gender			
Female	76 (41.3)	812 (47.7)	0.10
Male	108 (58.7)	892 (52.4)	

Characteristic	Children with OFC N = 184 (%)	Children without OFC N = 1,704 (%)	P-value
Child (cont.)			
Cleft Type			
Cleft lip	26 (14.1)	n/a	n/a
Cleft palate	67 (36.4)	n/a	
Cleft lip with cleft palate	91 (49.5)	n/a	
Presence of Other Anomalies[c]			
Isolated	123 (66.9)	n/a	n/a
Multiple	61 (33.2)	n/a	
System			
Source of Prenatal Care			
Health department	96 (52.2)	930 (54.6)	0.53
Other	88 (47.8)	774 (45.4)	
Receipt of Maternity Care Coordination Services			
Yes	86 (46.7)	913 (53.6)	0.08
No	98 (53.3)	791 (46.4)	
Receipt of WIC			
Yes	133 (72.3)	1,345 (78.9)	0.04*
No	51 (27.7)	359 (21.1)	
Birth Hospital Level of Care			
Level III	84 (45.7)	650 (38.2)	0.05
Community	100 (54.4)	1,054 (61.9)	
Perinatal Care Region			
Northwestern	49 (26.6)	389 (22.8)	0.21
Southwestern	29 (15.8)	224 (13.2)	
Northeastern	26 (14.1)	258 (15.1)	
Southeastern	26 (14.1)	330 (19.4)	
Eastern	32 (17.4)	353 (20.7)	
Western	22 (12.0)	150 (8.8)	
Place of Residence			
Metropolitan	113 (61.4)	998 (58.6)	0.68
Micropolitan	45 (24.5)	448 (26.3)	
Noncore areas adjacent to metro area or small town	19 (10.3)	163 (9.6)	
Noncore areas not adjacent to metro area or small town	7 (3.8)	95 (5.6)	

[a] Children were continuously enrolled in Medicaid during the fifth year of life
[b] Other includes Native American, Asian/Pacific Islander and other non-White
[c] Isolated anomaly = cleft lip with or without cleft palate diagnosis only diagnosis; multiple anomalies = cleft lip with or without cleft palate and other birth defect diagnosis
* Statistically significant ($p < 0.05$) by chi-square test

APPENDIX E: PATTERNS OF HEALTH SERVICE UTILIZATION AMONG CHILDREN WITH AND WITHOUT OROFACIAL CLEFTS AND BY CLEFT TYPE AND PRESENCE OF OTHER ANOMALIES AMONG CHILDREN WITH OROFACIAL CLEFTS DURING THE SECOND THROUGH FIFTH YEARS OF LIFE IN NORTH CAROLINA 1995-2002

Table 1. Mean Number of Medicaid Paid Claims and Range per Child for Children with and without Orofacial Clefts (OFC) during the 2nd Year of Life in North Carolina, 1995-2002

Service Category	Children with OFC (n = 406)		Children without OFC (n = 3,632)		Utilization Ratio[a]
	Mean	Range	Mean	Range	
Medical	43.83	1.00-267.00	31.03	0.00-211.00	1.41*
Inpatient	0.43	0.00-5.00	0.08	0.00-5.00	5.38*
Outpatient	3.67	0.00-49.00	1.56	0.00-31.00	2.35*
Mental Health	5.08	0.00-137.00	0.47	0.00-139.00	10.81*
Home Health	2.03	0.00-115.00	0.06	0.00-67.00	33.83*
Dental	0.22	0.00-8.00	0.05	0.00-3.00	4.40*
Well-child care	1.65	0.00-4.00	1.66	0.00-5.00	0.99
Other	0.11	0.00-4.00	0.04	0.005-5.00	2.75*
Total	57.02	1.00-365.00	34.93	1.00-252.00	1.63*

[a] Utilization ratio = ratio of mean number of paid claims per child with orofacial clefts to mean number of paid claims per child without orofacial clefts

* Utilization ratios for all categories of service except well-child care were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 2. Mean Number of Medicaid Paid Claims and Range per Child for Children with and without Orofacial Clefts (OFC) during the 3rd Year of Life in North Carolina, 1995-2002

Service Category	Children with OFC (n = 300)		Children without OFC (n = 2,708)		Utilization Ratio[a]
	Mean	Range	Mean	Range	
Medical	45.45	1.00-267.00	29.68	0.00-211.00	1.53*
Inpatient	0.20	0.00-7.00	0.04	0.00-5.00	5.00*
Outpatient	3.36	0.00-43.00	1.26	0.00-26.00	2.67*
Mental Health	7.72	0.00-137.00	1.07	0.00-139.00	7.21*
Home Health	1.89	0.00-79.00	0.03	0.00-31.00	63.00*
Dental	0.60	0.00-8.00	0.22	0.00-3.00	2.73*
Well-child care	0.71	0.00-3.00	0.68	0.00-4.00	1.04
Other	0.13	0.00-5.00	0.03	0.00-4.00	4.33*
Total	60.07	1.00-408.00	33.02	1.00-281.00	1.82*

[a] Utilization ratio = ratio of mean number of paid claims per child with orofacial clefts to mean number of paid claims per child without orofacial clefts

* Utilization ratios for all categories of service except well-child care were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 3. Mean Number of Medicaid Paid Claims and Range per Child for Children with and without Orofacial Clefts (OFC) during the 4th Year of Life in North Carolina, 1995-2002

Service Category	Children with OFC (n = 246)		Children without OFC (n = 2,135)		Utilization Ratio[a]
	Mean	Range	Mean	Range	
Medical	41.78	5.00-282.00	29.15	0.00-162.00	1.43*
Inpatient	0.18	0.00-7.00	0.03	0.00-3.00	6.00*
Outpatient	2.99	0.00-84.00	1.07	0.00-18.00	2.79*
Mental Health	2.63	0.00-169.00	0.71	0.00-249.00	3.70*
Home Health	1.16	0.00-69.00	0.05	0.00-84.00	23.20*
Dental	0.96	0.00-6.00	0.66	0.00-10.00	1.45*
Well-child care	0.59	0.00-2.00	0.60	0.00-3.00	0.98
Other	0.15	0.00-3.00	0.06	0.00-11.00	2.50*
Total	50.43	6.00-330.00	32.32	1.00-361.00	1.56*

[a] Utilization ratio = ratio of mean number of paid claims per child with orofacial clefts to mean number of paid claims per child without orofacial clefts

* Utilization ratios for all categories of service except well-child care were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 4. Mean Number of Medicaid Paid Claims and Range per Child for Children with and without Orofacial Clefts (OFC) during the 5th Year of Life in North Carolina, 1995-2002

Service Category	Children with OFC (n = 184)		Children without OFC (n = 1,704)		Utilization Ratio[a]
	Mean	Range	Mean	Range	
Medical	37.75	1.00-144.00	29.69	0.00-188.00	1.27*
Inpatient	0.07	0.00-3.00	0.03	0.00-3.00	2.33*
Outpatient	2.65	0.00-60.00	0.99	0.00-22.00	2.68*
Mental Health	2.99	0.00-151.00	0.60	0.00-224.00	4.98*
Home Health	0.72	0.00-47.00	0.04	0.00-43.00	18.00*
Dental	1.15	0.00-6.00	0.98	0.00-10.00	1.17*
Well-child care	0.71	0.00-2.00	0.74	0.00-5.00	0.96
Other	0.14	0.00-5.00	0.08	0.00-8.00	1.75
Total	46.19	1.00-181.00	33.17	1.00-301.00	1.39*

[a] Utilization ratio = ratio of mean number of paid claims per child with orofacial clefts to mean number of paid claims per child without orofacial clefts

* Utilization ratios for all categories of service except well-child care and other were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 5. Mean Number of Medicaid Paid Claims per Child by Cleft Type and Year of Life for Children with Orofacial Clefts in North Carolina, 1995-2002

Service Category	Cleft Type[a]	Year of Life			
		2	3	4	5
Medical	CP	45.53	46.30	43.17	37.81
	CL	33.86	32.57	34.64	31.00
	CLP	46.47	48.85	42.86	39.64
Inpatient	CP	0.57	0.16	0.27	0.06
	CL	0.12	0.05	0.03	0.04
	CLP	0.43	0.28	0.16	0.09
Outpatient	CP	4.03	3.35	3.62	3.27
	CL	2.14	1.36	1.47	0.77
	CLP	4.01	4.01	2.96	2.74
Mental Health	CP	5.09	6.86	4.05	3.28
	CL	3.46	6.41	0.86	2.77
	CLP	5.73	8.89	2.02	2.85
Home Health	CP	2.33	1.87	1.66	1.01
	CL	0.00	0.00	0.00	0.00
	CLP	2.60	2.53	1.11	0.71
Dental	CP	0.19	0.58	0.99	1.21
	CL	0.14	0.39	0.78	0.85
	CLP	0.27	0.69	0.99	1.20
Well-child Care	CP	1.70	0.74	0.62	0.70
	CL	1.49	0.70	0.53	0.62
	CLP	1.68	0.69	0.59	0.75
Other	CP	0.13	0.13	0.10	0.09
	CL	0.08	0.09	0.17	0.08
	CLP	0.12	0.15	0.18	0.19
Total	CP	59.58	59.98	54.47	47.43
	CL	41.28	41.57	38.47	36.12
	CLP	61.31	66.08	50.86	48.15

[a] CP = cleft palate only; CL = cleft lip only; CLP = cleft lip with cleft palate

Table 6. Mean Number of Medicaid Paid Claims per Child by Presence of Other Anomalies and Year of Life for Children with Orofacial Clefts in North Carolina, 1995-2002

Service Category	Presence of Other Anomalies[a]	Year of Life			
		2	3	4	5
Medical	Isolated Anomaly	35.40	35.87	36.54	34.62
	Multiple Anomalies	58.84	60.03	51.02	44.07
Inpatient	Isolated Anomaly	0.28	0.07	0.10	0.04
	Multiple Anomalies	0.69	0.39	0.31	0.13
Outpatient	Isolated Anomaly	2.44	2.15	2.25	1.89
	Multiple Anomalies	5.87	5.19	4.29	4.20
Mental Health	Isolated Anomaly	2.97	5.85	1.59	1.85
	Multiple Anomalies	8.83	10.56	4.45	5.31
Home Health	Isolated Anomaly	0.10	0.05	0.01	0.02
	Multiple Anomalies	5.47	4.70	3.18	2.15
Dental	Isolated Anomaly	0.22	0.56	0.92	1.04
	Multiple Anomalies	0.21	0.66	1.03	1.38
Well-child Care	Isolated Anomaly	1.61	0.67	0.61	0.78
	Multiple Anomalies	1.73	0.77	0.55	0.57
Other	Isolated Anomaly	0.04	0.08	0.08	0.02
	Multiple Anomalies	0.24	0.20	0.27	0.36
Total	Isolated Anomaly	43.06	45.31	42.10	40.25
	Multiple Anomalies	81.88	82.51	65.11	58.16

[a] Isolated anomaly = cleft lip with or without cleft palate diagnosis only diagnosis; multiple anomalies = cleft lip with or without cleft palate and other birth defect diagnosis

APPENDIX F: PATTERNS OF HEALTH COSTS AMONG CHILDREN WITH AND WITHOUT OROFACIAL CLEFTS AND BY CLEFT TYPE AND PRESENCE OF OTHER ANOMALIES AMONG CHILDREN WITH OROFACIAL CLEFTS DURING THE SECOND THROUGH FIFTH YEAR OF LIFE IN NORTH CAROLINA, 1995-2002

Table 1. Mean Cost and Range in Dollars per Child for Children with and without Orofacial Clefts (OFC) during the 2nd Year of Life in North Carolina, 1995-2002

Cost Category	Children with OFC (n = 406)		Children without OFC (n = 3632)		Cost Ratio[a]
	Mean	Range	Mean	Range	
Medical	3,992	27-212,558	651	0-23,208	6.13*
Inpatient	2,434	0-99,910	186	0-39,960	13.09*
Outpatient	1,740	0-21,487	229	0-13,575	7.60*
Mental Health	1,137	0-129,528	74	0-17,797	15.36*
Home Health	4,086	0-196,135	46	0-84,841	88.83*
Dental	23	0-1,547	7	0-1,318	3.29*
Well-child Care	126	0-321	129	0-416	0.98
Other	11	0-1,333	4	0-2,026	2.75*
Total	13,549	32-284,858	1,326	0-109,224	10.22*

[a] Cost ratio = ratio of mean cost per child with orofacial clefts to mean cost per child without orofacial clefts

* Cost ratios for all cost categories except well-child care were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 2. Mean Cost and Range in Dollars per Child for Children with and without Orofacial Clefts (OFC) during the 3rd Year of Life in North Carolina, 1995-2002

Cost Category	Children with OFC (n = 300)		Children without OFC (n = 2,708)		Cost Ratio[a]
	Mean	Range	Mean	Range	
Medical	5,044	0-238,921	606	0-27,533	8.32*
Inpatient	1,650	0-139,456	187	0-246,848	8.82*
Outpatient	1,274	0-335,394	207	0-9,019	6.15*
Mental Health	1,411	0-136,837	156	0-13,460	9.04*
Home Health	3,523	0-200,957	44	0-72,402	80.07*
Dental	106	0-2,668	37	0-3,518	2.86*
Well-child Care	55	0-234	54	0-321	1.02
Other	9	0-794	3	0-674	3.00*
Total	13,071	0-291,772	1,294	0-347,643	10.10*

[a] Cost ratio = ratio of mean cost per child with orofacial clefts to mean cost per child without orofacial clefts

* Cost ratios for all cost categories except well-child care were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 3. Mean Cost and Range in Dollars per Child for Children with and without Orofacial Clefts (OFC) during the 4th Year of Life in North Carolina, 1995-2002

Cost Category	Children with OFC (n = 246)		Children without OFC (n = 2135)		Cost Ratio[a]
	Mean	Range	Mean	Range	
Medical	3,079	2-183,679	563	0-30,485	5.47*
Inpatient	1,014	0-66,081	129	0-83,355	7.86*
Outpatient	1,051	0-19,632	193	0-6,418	5.45*
Mental Health	957	0-139,593	113	0-32,985	8.47*
Home Health	1,709	0-190,414	111	0-224,281	15.40*
Dental	164	0-2,372	103	0-3,604	1.59*
Well-child Care	46	0-180	48	0-251	0.96
Other	8	0-599	3	0-1,628	2.67*
Total	8,658	5-227,546	1,264	0-257,784	6.85*

[a] Cost ratio = ratio of mean cost per child with orofacial clefts to mean cost per child without orofacial clefts

* Cost ratios for all cost categories except well-child care were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 4. Mean Cost and Range in Dollars per Child for Children with and without Orofacial Clefts (OFC) during the 5th Year of Life in North Carolina, 1995-2002

Cost Category	Children with OFC (n = 184)		Children without OFC (n = 1704)		Cost Ratio[a]
	Mean	Range	Mean	Range	
Medical	2,809	1-139,733	552	0-56,235	5.09*
Inpatient	421	0-28,414	149	0-75,415	2.83*
Outpatient	1,155	0-20,004	194	0-18,269	5.95*
Mental Health	1,447	0-142,938	162	0-71,265	8.93*
Home Health	1,317	0-198,706	130	0-211,691	10.13*
Dental	182	0-2,059	154	0-4,232	1.18*
Well-child Care	56	0-180	59	0-374	0.95
Other	12	0-728	5	0-2,247	2.40
Total	7,399	1-234,572	1,406	0-345,080	5.26*

[a] Cost ratio = ratio of mean cost per child with orofacial clefts to mean cost per child without orofacial clefts

* Cost ratios for all cost categories except well-child care and other were statistically significant ($p < 0.05$) by Wilcoxon rank sum test

Table 5. Mean Cost in Dollars per Child by Cleft Type and Year of Life for Children with Orofacial Clefts in North Carolina, 1995-2002

Cost Category	Cleft Type[a]	Year of Life			
		2	**3**	**4**	**5**
Medical	CP	4,544	4,751	3,301	2,447
	CL	1,083	1,150	867	836
	CLP	4,719	6,548	4,921	3,639
Inpatient	CP	3,108	2,075	1,440	285
	CL	279	440	42	43
	CLP	2,754	1,668	971	630
Outpatient	CP	1,408	1,240	1,198	1,193
	CL	605	556	374	506
	CLP	2,476	1,535	1,143	1,312
Mental Health	CP	2,056	1,938	2,008	3,628
	CL	260	644	274	226
	CLP	737	1,200	318	190
Home Health	CP	4,410	3,627	3,343	3,094
	CL	0	0	0	0
	CLP	5,481	4,564	915	385
Dental	CP	27	103	194	201
	CL	5	92	123	88
	CLP	26	112	151	195
Well-child care	CP	130	57	48	57
	CL	116	54	41	47
	CLP	128	53	46	58
Other	CP	16	9	8	10
	CL	5	2	5	2
	CLP	9	12	8	16
Total	CP	15,699	13,800	11,540	10,915
	CL	2,353	2,938	1,725	1,750
	CLP	16,330	15,693	8,474	6,424

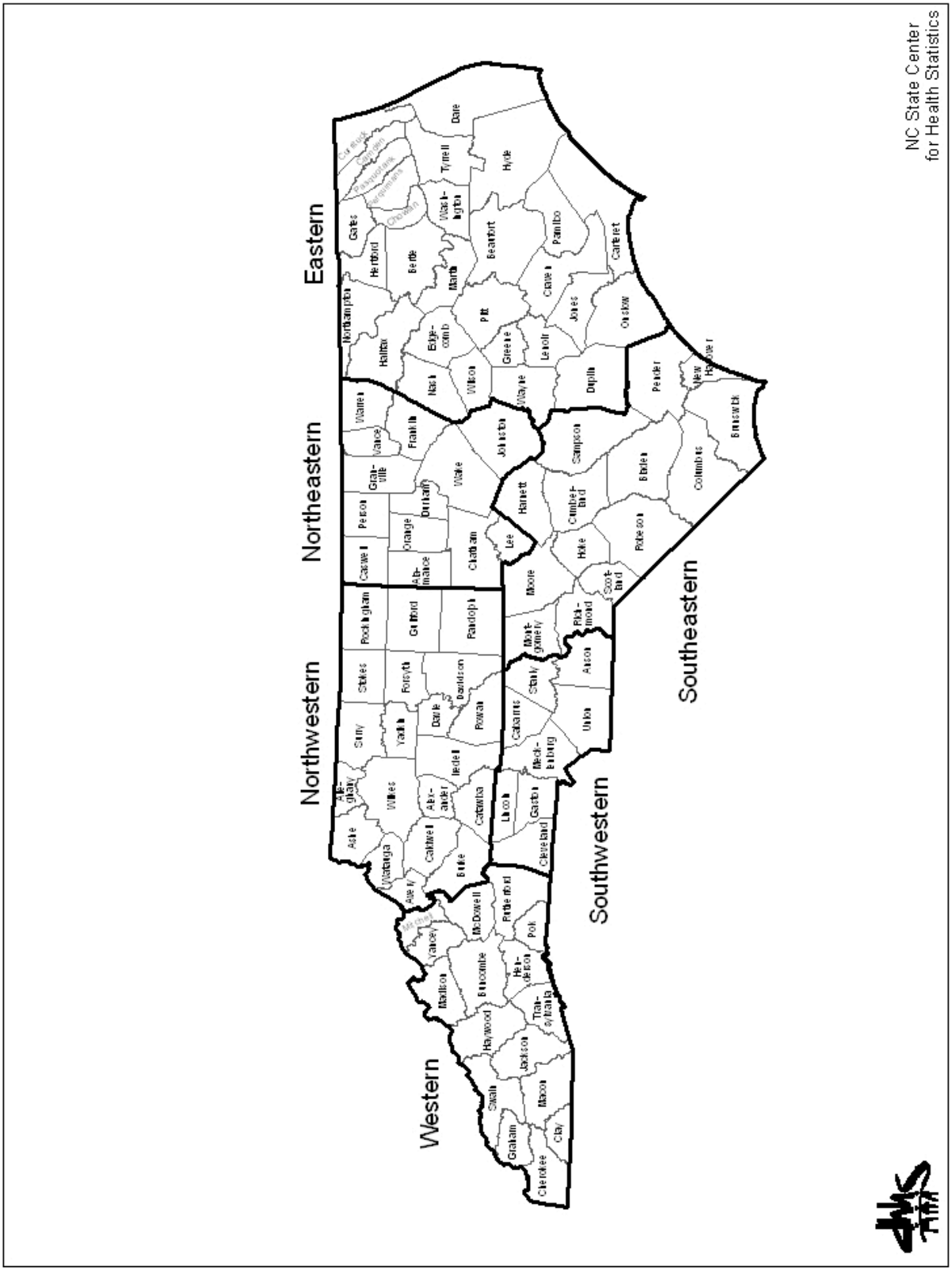
[a] CP = cleft palate only; CL = cleft lip only; CLP = cleft lip with cleft palate

Table 6. Mean Cost in Dollars per Child by Presence of Other Anomalies and Year of Life for Children with Orofacial Clefts in North Carolina, 1995-2002

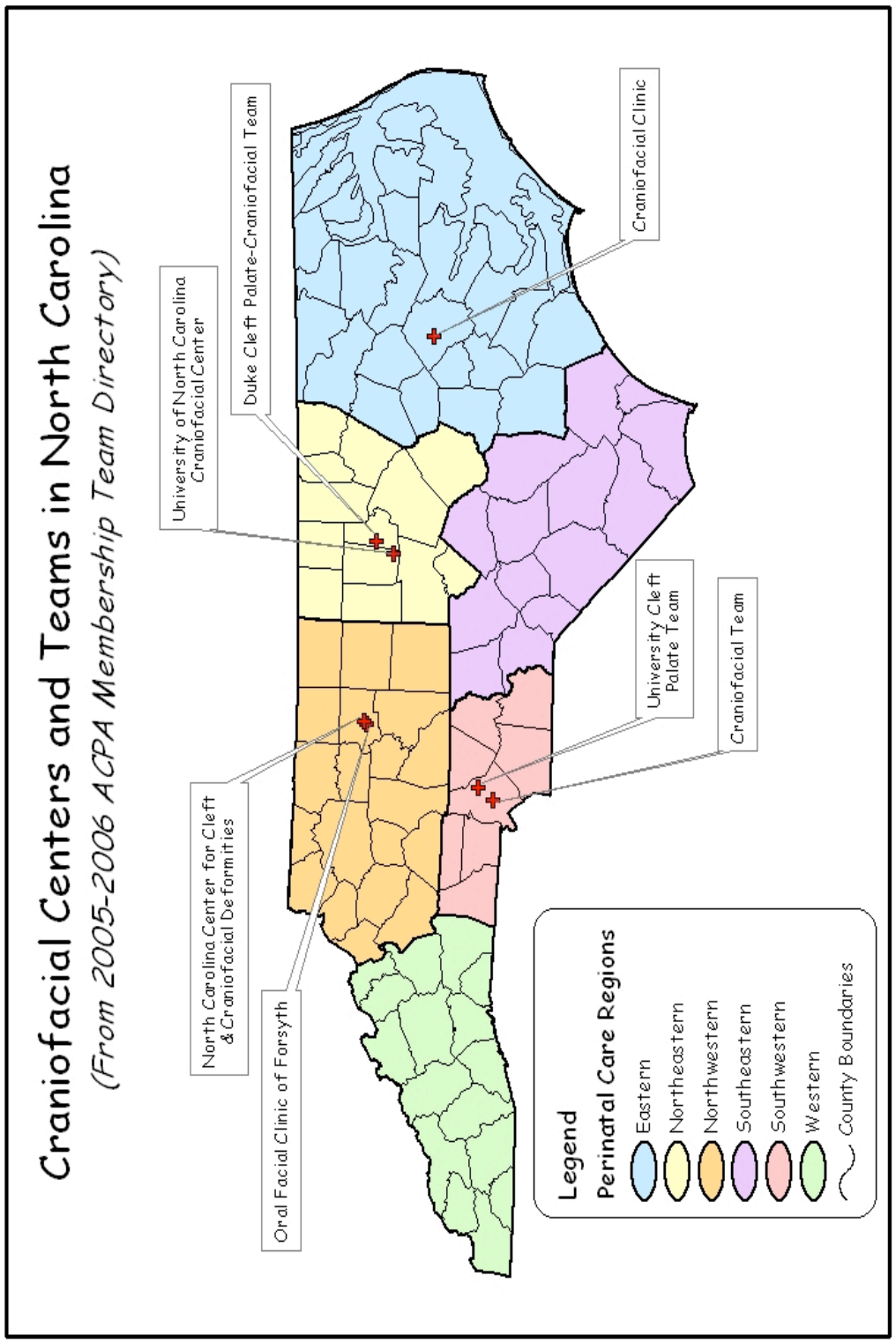
Cost Category	Presence of Other Anomalies[a]	Year of Life			
		2	**3**	**4**	**5**
Medical	Isolated Anomaly	1,395	1,456	1,581	1,434
	Multiple Anomalies	8,617	10,501	7,463	5,581
Inpatient	Isolated Anomaly	912	317	462	173
	Multiple Anomalies	5,144	3,676	1,988	923
Outpatient	Isolated Anomaly	1,345	821	823	743
	Multiple Anomalies	2,444	1,964	1,455	1,985
Mental Health	Isolated Anomaly	376	803	367	230
	Multiple Anomalies	2,493	2,335	1,998	3,902
Home Health	Isolated Anomaly	29	16	3	4
	Multiple Anomalies	11,311	8,856	4,719	3,966
Dental	Isolated Anomaly	22	84	180	137
	Multiple Anomalies	23	139	134	274
Well-child care	Isolated Anomaly	124	52	49	61
	Multiple Anomalies	130	60	41	46
Other	Isolated Anomaly	3	4	3	0
	Multiple Anomalies	25	17	17	34
Total	Isolated Anomaly	4,207	3,554	3,648	2,781
	Multiple Anomalies	30,187	27,547	17,814	16,711

[a] Isolated anomaly = cleft lip with or without cleft palate diagnosis only; multiple anomalies = cleft lip with or without cleft palate and other birth defect diagnosis

APPENDIX G: MAP OF PERINATAL CARE REGIONS IN NORTH CAROLINA



APPENDIX H: MAP OF CRANIOFACIAL CENTERS AND TEAMS IN NORTH CAROLINA



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