

Treatment of Oralfacial Clefts by State-affiliated Craniofacial Centers and Cleft Palate Clinics

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BACKGROUND: Oralfacial clefting (OFC) disorders require expedient evaluation and treatment to obtain optimal outcome. In Florida, there is a statewide program targeted to the care of infants with OFC. We therefore sought to determine statewide referral and treatment patterns of children born with OFC identified through the Florida Birth Defects Registry. **METHODS:** Using data for 1996 and 1997 and ICD-9 CM codes 749.00 - 749.25, we identified 539 OFC cases. All cases were matched with the evaluation and treatment records of the statewide Children's Medical Services' (CMS) craniofacial centers (CFC) and cleft palate clinics (CPC). The likelihood of CMS contact was examined with respect to demographic and other descriptive data characterizing the OFC cases. **RESULTS:** 42% (227/539) of OFC cases were evaluated at or known to the CFC or CPC. Children with cleft lip and palate were more likely to have had contact than were those with cleft lip or cleft palate alone. The CFC and CPC programs were most likely to provide evaluation between age 2 months and 3 years. Of 12 counties with occurrences of more than 15 OFC cases, 2 had significantly lower contact rates, suggesting possible problems in accessibility or reporting of services. **CONCLUSIONS:** Statewide Birth Defect Registry data can be used in collaboration with statewide treatment programs to gain insight into referral patterns and provision of services. Factors influencing access to services and quality of care, though not addressed by this study, could be prospectively incorporated into such a project. *Birth Defects Research (Part A) 67:643–646, 2003.* © 2003 Wiley-Liss, Inc.

INTRODUCTION

Birth defect registry data are increasingly being used to link the provision of health services to specific birth defects. For newborns with oralfacial clefting (OFC), the use of registries could be important since OFC is readily diagnosed, and expedient provision of multidisciplinary care has been accepted as an ideal approach to treatment. The state of Florida has established multidisciplinary craniofacial clinics and smaller cleft palate clinics throughout the state to better coordinate such care. To assist the state in understanding the scope and effectiveness of these clinics, we sought to track newborns with OFC to determine statewide referral and treatment patterns.

MATERIALS AND METHODS

The Florida Birth Defects Registry (FBDR) data was based on surveillance from birth to age one year of all infants born in Florida (from January 1, 1996 to December 31, 1997) with ICD-9 CM codes that described a cleft of the lip or palate or both (749.0 to 749.25). The Registry uses multiple sources to identify cases, the primary sources being the Florida Department of Health Office of Vital Statistics, Children's Medical Services program, and the Florida Agency for Health Care Administration hospital discharge database. Cases include infants up to one year of

age born to Florida residents. Maternal information, such as county of residence, ethnicity, and place of birth came from the birth certificate.

The FBDR dataset was imported into a Microsoft Access database designed to facilitate review and categorization of the cases. The ICD-9 CM codes and the sources of the codes used to ascertain the OFC case were viewed concurrently and reconciled, by a clinical geneticist. For example, in cases where ICD-9 CM codes differed for severity or type of clefting, the more extensive malformation was selected as the "correct" description (e.g., where a case was coded for cleft lip alone in one data source but was coded for cleft lip and palate in another). For each OFC case, all other reported ICD-9 CM codes were also viewed to determine whether the case was associated with chromosome or teratogen-related causation, a known multiple congenital anomaly (MCA) syndrome or a sequence, or associated with multiple anomalies of unknown etiology or syndromic designation.

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Table 1
Contact with CMS by Maternal Age, Race
and Ethnicity

	CMS contact		Total cases	P value*
	No	Yes		
Maternal age				
<20	31 (43%)	41 (57%)	72	.40
20–24	62 (48%)	66 (52%)	128	.77
25–29	88 (63%)	51 (37%)	139	.025
30–34	78 (63%)	45 (37%)	123	.034
35+	53 (69%)	24 (31%)	77	.017
Maternal Race				
White	254 (58%)	183 (42%)	437	.016
Black	43 (53%)	38 (47%)	81	.70
Asian/Pacific Islander	13 (72%)	5 (28%)	18	
Native American	2 (67%)	1 (33%)	3	
Hispanic Ethnicity				
Gen. or South American	20 (71%)	8 (29%)	28	
Mexican	4 (21%)	15 (79%)	19	
Puerto Rican	3 (21%)	11 (79%)	14	
Cuban	11 (85%)	2 (15%)	13	
Haitian	4 (44%)	5 (56%)	9	
Other Hispanic	2 (29%)	5 (71%)	7	
Total Hispanic	44 (49%)	46 (51%)	90	.88
Non-Hispanic	268 (60%)	181 (40%)	449	
Total	312 (58%)	227 (42%)	539	

*Chi-square.

All OFC cases were subsequently matched with evaluation and treatment records of the statewide Children's Medical Services' craniofacial centers (CFC) and cleft palate clinics (CPC), using a state-approved research protocol. Treatment and follow-up information were obtained by review of CMS centralized electronic databases, treatment rosters at the respective centers, and patient medical records. Data obtained included demographic information, contact status (e.g., current patient, closed case, etc.), OFC ICD-9 CM codes, date of first evaluation, date and type of surgical procedures, surgical treatment facilities, and follow-up clinic sites. This information was entered into an Excel spreadsheet, then imported into the Microsoft Access database. Comparisons were then made between the FBDR-identified OFC cases having contact and those not having contact with these clinics. Chi-square and odds ratio analyses were used for statistical evaluations.

RESULTS

OFC Descriptive Data

There were 381,128 live births during the period of review; 539 live born OFC cases were identified giving a birth prevalence of 1.41 per 1,000. Of these cases, 73.3% had ICD-9 CM codes relating only to clefting and these cases were presumed to have a single and not a multiple birth defect. Of the 539 OFC cases, 45.5% had cleft lip (CL) with cleft palate (CP), 16% had CL alone and 38.5% had CP. Further review of these cases revealed 9.6% having an apparent identified causation or pattern of anomalies (6.3% chromosomal, 2.2% teratogen, 0.7% known MCA, and 0.4% sequence), while 90.3% were of unknown etiology (18.7% had multiple malformations and 71.6% had the OFC as the only reported birth defect). Male to female

ratios for types of clefting were 1.49, 1.39 and 1.14 for CLP, CL and CP respectively. Oralfacial clefting was the only reported birth defect in 75%, 90.7% and 58.2% of cases with CLP, CL and CP respectively. Rates of clefting for all types of OFCs were higher in whites (1.52/1,000) than in Black (0.95/1,000; O.R. 0.67 [0.53–0.85]) and Hispanic categories (.98/1,000; O.R. 0.68[0.51–0.82]). Similar trends were observed for CL with or without CP, and for CP alone, in these three racial/ethnic categories (data not shown).

OFC Treatment and Evaluation Data

Cases that were known to have had contact with the Florida's CMS-sponsored CFC or CPC represented 42.1% (227/539) of all OFC cases reported to the Florida registry. Fifty-two percent of all cases with cleft lip with cleft palate had CMS contact compared to 38% with CP alone and 24% with CL alone. Of the 227 children with CMS contact, 52% had at least one visit at a CPC and the statewide CFCs had contact with 31.6% of cases. Table 1 depicts maternal age, race and ethnicity in relation to CMS contact status. Mothers 25–29, 30–34, and >35 years of age were significantly less likely to have had CMS contact than were mothers under age 25 years. As a group, mothers less than age 25 years were about twice as likely to have had CMS contact than those over age 25 years or older (OR: 2.10 95% CI: 1.47, 3.00). There was no significant difference in the CMS contact rate when the mother was either Black or Hispanic, but Whites were significantly less likely to have had contact. Ninety-five percent of cases with contact were first seen when they were less than age 3 years, and of these, 26% had their first contact at age less than two months. Cases with only CL were significantly less likely to have any contact with either a CFC or CPC (see table 2).

Of the state's counties with occurrences of more than 15 OFC cases, 2 had markedly lower CFC and CPC contact rates (see table 3). Figure 1 indicates the distribution of all OFC cases and their CMS contact status. Geographic occurrences reflected areas of population density.

DISCUSSION

The main objective of our study was to determine to what extent infants with OFC were evaluated and treated by the State of Florida's CMS program. Legislative action in Florida in the last decade had established insurance guidelines for comprehensive treatment of OFC and the state had invested substantially in regional CFCs and CPCs. Surgical repair of OFC is often complicated (Kirsch-

Table 2
CMS Contact by Cleft Type and Severity

Cleft type	CMS contact*		Total	P**
	No	Yes		
Cleft Lip	65 (76%)	21 (24%)	86	<.001
Cleft Palate	130 (62%)	78 (38%)	208	.01
Cleft Lip and Palate	117 (48%)	128 (52%)	245	.62
Total	312 (58%)	227 (42%)	539	539

*CMS Contact includes: Current CMS patient, current private patient with previous CMS evaluation, transfer to other CMS location, CMS monitoring/reimbursement only with no CMS evaluation, CMS closed case, and other.

**Chi-square.

Table 3
CMS Contact by County of Residence (Counties with
15 or more Patients)

County	CMS contact*		Total cases
	No	Yes	
Miami-Dade	52 (83%)	11 (17%)	63
Palm Beach	32 (67%)	16 (33%)	48
Hillsborough	16 (44%)	20 (56%)	36
Orange	25 (71%)	10 (29%)	35
Broward	25 (74%)	9 (26%)	34
Pinellas	17 (63%)	10 (37%)	27
Duval	16 (70%)	7 (30%)	23
Lee	7 (32%)	15 (68%)	22
Brevard	17 (89%)	2 (11%)	19
Polk	6 (32%)	13 (68%)	19
Marion	3 (20%)	12 (80%)	15
Volusia	6 (40%)	9 (60%)	15
Other and Unknown	90 (49%)	93 (51%)	183
Total	312 (58%)	227 (42%)	539

*CMS Contact includes: Current CMS patient, current private patient with previous CMS evaluation, transfer to other CMS location, CMS monitoring/reimbursement only with no CMS evaluation, CMS closed case, and other.

ner and LaRossa, '00) and these clinics are therefore intended to promote the recognized goals of early evaluation and multidisciplinary care (Habel et al., '96; Thomas, '00). Since medical care for the treatment of OFC can be secured in the private sector, it was expected that a large percent of all cases would not have had evaluation with the state CFC or CPC programs. Indeed we found that only 42% of cases

had any contact with these CMS programs. We were surprised to find that mothers less than 25 years of age were more likely to have had contact with the CMS program than were those age 25 years or older. We did not have availability of maternal socio-economic information but it is our speculation that the financial capability of the mother may explain a large part of this phenomenon.

More important in our study was the pattern of reported statewide use of the CFC or CPC. Of twelve counties that had more than 15 cases of OFC during the reporting period, we identified two that had apparent low contact rates of 11% and 17% (see table 2). Although the state may further review this finding, our study methods could not identify whether these two counties were underreporting the contact rate, whether they have under-utilization of the CFCs and CPCs, or whether there are more private treatment alternatives in these areas. We also found that infants with more complicated malformations were more likely to be seen in the CFC and CPC programs (see table 1).

The descriptive findings that we observed herein for OFCs are generally similar to those reported in a recent, epidemiological study conducted in California (Tolarova and Cervenka, '98). In our study, OFC had a relatively common birth prevalence of 1.41/1,000. Cleft lip with or without cleft palate was more common in males and had significantly higher prevalence in non-Hispanic Whites. Most OFC malformations did not have a known etiology but were more likely to occur as an isolated malformation when the OFC was the CL±CP type rather than CP alone.

Although our study was retrospective, there is increasing interest in the use of Birth Defect Registries for more contemporaneous linking of services to certain defects,

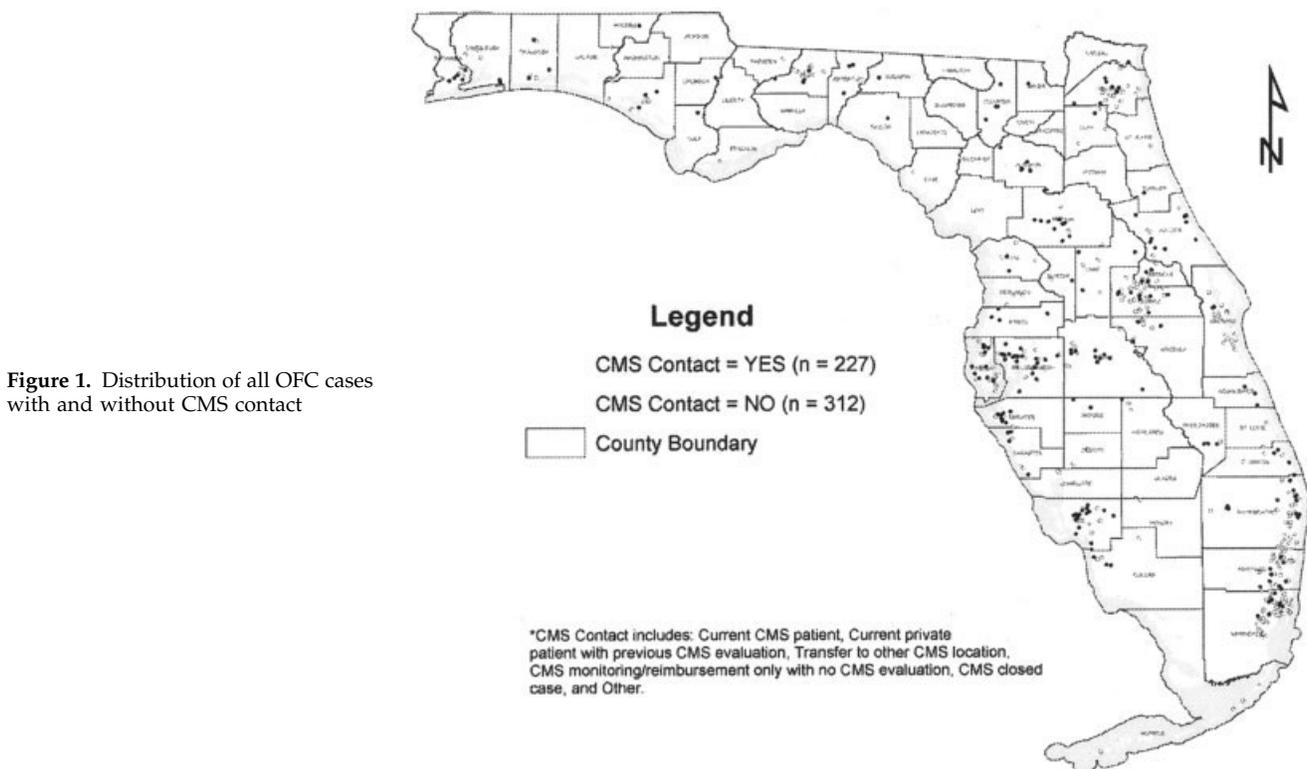


Figure 1. Distribution of all OFC cases with and without CMS contact

especially those associated with developmental handicap (Montgomery and Miller, '01). The results of our study have helped further discussions within the state about how linkages with the registry may improve access to evaluation and treatment programs. To facilitate use of our findings, we presented the study results at a statewide Birth Defects Advisory Committee meeting and at the statewide Craniofacial and Cleft Palate Association meeting.

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