The Need for Orthognathic Surgery in Patients With Repaired Complete Unilateral and Complete Bilateral Cleft Lip and Palate

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Objective: **To determine the percentage of patients with complete unilateral cleft lip and palate and complete bilateral cleft lip and palate treated at SickKids since birth who would benefit from orthognathic surgery.**

Design: **Retrospective cohort study.**

Subjects: **The review comprised records of 258 patients with complete unilateral cleft lip and palate and 149 patients with complete bilateral cleft lip and palate born from 1960 to 1989. Of these, 211 and 129 patients, respectively, had been treated at SickKids since birth. Patients with syndromes or associated anomalies were excluded.**

Methods: **Patients who had undergone orthognathic surgery were recorded. For the remaining patients, arbitrarily set cephalometric criteria were used in order to identify the ''objective'' need for surgery. Lateral cephalometric radiographs taken beyond the age of 15 years were digitized using Dentofacial Planner cephalometric software.**

Results: **Of the 211 patients with complete unilateral cleft lip and palate, 102 (48.3%) were deemed to benefit from orthognathic surgery. For the complete bilateral cleft lip and palate sample, the percentage was 65.1% (84 of 129). Definitive information on presurgical orthopedics was available for a small subsample (101 patients) of the complete unilateral cleft lip and palate cohort. The need for orthognathic surgery for this group was slightly higher (59.4%, or 60 of 101).**

Conclusion: **These results suggest that a considerable percentage of patients with a history of complete cleft lip and palate at our institution require orthognathic surgery. Factors that need to be considered in the interpretation of these results include the quest for improvement in the profile aesthetics; the fact that the Canadian health care system covers the costs of surgery, making it more accessible to the patients; and the inclusion in the above figures of patients who had orthognathic surgery solely for reasons of closure of previously ungrafted alveolar clefts and associated fistulae.**

KEY WORDS: *complete cleft lip and palate, frequency, orthognathic surgery*

Patients born with cleft lip and palate (CLP) must undergo a number of corrective surgical procedures during their infancy and early childhood. The scarring that results from these procedures has been shown to affect the growth of the maxilla, often leading to maxillary deficiency (Ross, 1987; Mars et al., 1990; Capelozza Filho et al., 1996). The timing and execution of initial repairs and subsequent interventions have been scrutinized and refined in an attempt to maximize the benefits while keeping the interference with maxillary growth to a minimum. Nevertheless, a percentage of patients with CLP go on to require orthognathic surgery at skeletal maturity for correction of a skeletal class III malocclusion.

The frequency of need for orthognathic surgery carries particular significance for a specific institution because it is generally assumed to reflect the success or failure of the institution's treatment protocol. It is, therefore, not surprising that there are remarkably few reports of orthognathic surgery rates for patients with CLP in the literature.

Ross (1986) estimated that orthognathic surgery would be necessary in approximately 25% of a sample of men with unilateral cleft lip and palate (UCLP) to permit adequate functional jaw relations, harmonious facial aesthetics, or both. Along the same lines, DeLuke et al. (1997) reported that 25% of 28 patients with mixed types of CLP required orthognathic surgery, having followed their institution's

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treatment protocol. On the other hand, Rosenstein et al. (2003) stated that in their center, where primary bone grafting is performed at the time of initial lip repair, the rate of orthognathic surgery was 18.29% in a sample of 82 patients with mixed types of CLP.

In another report by Schnitt et al. (2004), 32% (7 of 22) of patients with UCLP from the Australian Craniofacial Unit in Adelaide were found retrospectively to have required maxillary or bimaxillary surgery for their eventual rehabilitation. Similarly, Cohen et al. (1995) recommended orthognathic surgery to 26% (10 of 38) of patients with UCLP and 24% (7 of 29) of patients with bilateral cleft lip and palate (BCLP), respectively, who were consecutively treated in their institution.

Significant variation in the percentage of patients with complete UCLP who were deemed to require an osteotomy was reported in the Eurocleft study (Mølsted et al., 2005). The rate of orthognathic surgery in the five European centers involved was 4%, 7%, 17%, 45%, and 50%, respectively.

More recently, a group from Boston Children's Hospital reported frequencies of Le Fort I osteotomy of 48.5% (16 out of 33) for patients with complete UCLP and 76.5% (13 out of 17) for patients with complete BCLP treated by a single surgeon (Good et al., 2007).

The purpose of this retrospective study was to determine the frequency of need for orthognathic surgery among patients with a history of UCLP and BCLP at SickKids Hospital in Toronto, Ontario, Canada.

Subjects and Methods

Following approval by the SickKids Research Ethics Board, a review of the databases of the Cleft Lip and Palate Programme and the Orthodontic Clinic was performed, with reference to the initial diagnosis (type of cleft, accompanying anomalies) and the records available. Those selected were all patients with either complete unilateral (CUCLP) or complete bilateral (CBCLP) cleft lip and palate born between 1960 and 1989 whose records were available. The required records were photographs at initial presentation and/or an operative report describing the extent of the initial cleft condition, as well as a lateral cephalometric radiograph taken at a minimum age of 15 years. This was considered to be an age where one could make a reasonable judgment as to the eventual need (or not) for orthognathic surgery due to a skeletal malrelationship. Patients with Simonart bands at initial presentation were included in the sample. Patients with incomplete clefts or with associated anomalies at the original presentation or patients who had initial repairs performed at other institutions were excluded.

Those who had undergone orthognathic surgery or distraction osteogenesis or were in the process of presurgical orthodontic preparation were automatically classified as "needing surgery." For the remaining patients, the most recent cephalometric radiographs available (minimum 15 years) were traced and analyzed using Dentofacial Planner version 7.0 computer software (Dentofacial Software, Toronto, ON, Canada). An objective determination of the need for orthognathic surgery was made based on the satisfaction of all three of the following criteria (which were selected arbitrarily): ANB angle of −3 degrees or lower; Harvold unit difference (CoGn-CoSn') of 34 mm or larger; and Wits appraisal result of −5 mm or lower (B-point ahead of A-point) (Linton, 1998). Patients who fulfilled all three of the above criteria were deemed to be "objective" orthognathic candidates, irrespective of the fact that no surgery had actually been performed on them. Other variables recorded (when available) were surgeon performing the initial repair and whether presurgical orthopedics was performed prior to the primary repair.

TABLE 1 Samples Examined in the Study

	*CUCLP**			*CBCLP*		
	SickKids	*Non-SickKids*	*Total*	*SickKids*	*Non-SickKids*	*Total*
Male	126	26	152	91	17	108
Female	85	21	106	38	3	41
Male + female	211	47	258	129	20	149

* CUCLP = complete unilateral cleft lip and palate; CBCLP = complete bilateral cleft lip and palate.

Results

Complete Unilateral Cleft Lip and Palate

The number of patients with CUCLP who satisfied the inclusion criteria in terms of type of cleft and available records totaled 258. Of these, 211 patients had been treated at SickKids since birth. Of the SickKids sample, 28% (60 patients) had undergone orthognathic surgery or were under presurgical orthodontic preparation at the time of review of the records. When the remaining patients were evaluated based on the objective cephalometric criteria outlined earlier, the percentage of patients with CUCLP judged to require orthognathic surgery rose to 48.3% (Table 1). When looking at the data by decade, a trend was observed: The need for surgery increased for patients born in the 1980s compared with those born in the 1960s.

Complete Bilateral Cleft Lip and Palate

The number of patients with CBCLP who satisfied the inclusion criteria in terms of type of cleft and available records totaled 149. Of these, 129 had been treated at SickKids since birth. Of the SickKids sample, 38% (49 patients) had undergone orthognathic surgery already or were under presurgical orthodontic preparation at the time of review. When the objective cephalometric criteria were applied to the remaining patients, the percentage of patients with CUCLP judged to require orthognathic surgery rose to 65.1% (Table 1). Similarly to the CUCLP

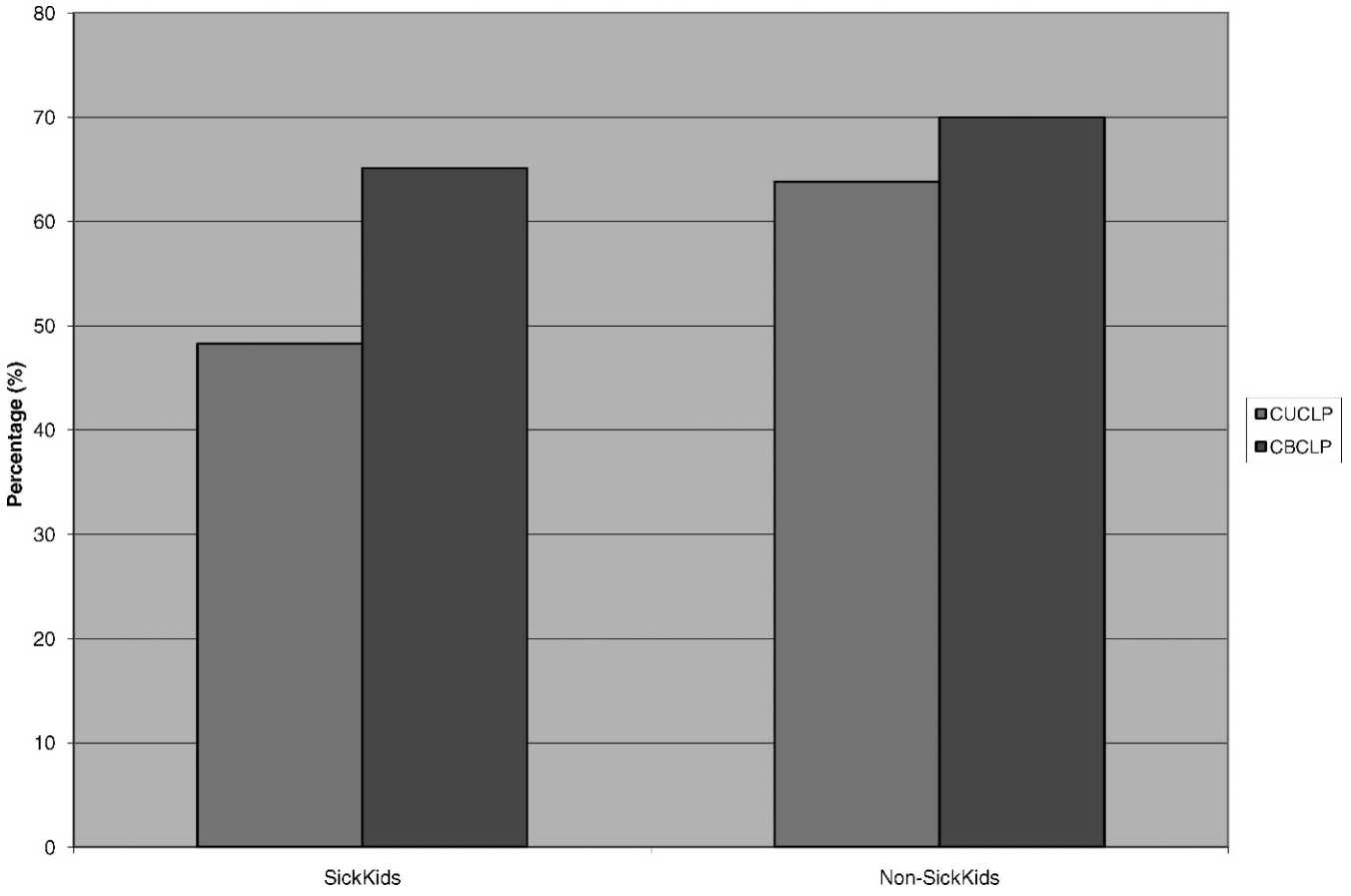

FIGURE 1 Percentages of patients deemed to require orthognathic surgery, by the type of cleft and institution of initial cleft repair.

data, a trend of increasing need for surgery by decade of birth was noted.

With regard to the presurgical orthopedics, insufficient information was available for the majority of the samples. In the CUCLP sample, 101 patients were clearly documented to have received presurgical orthopedics prior to their lip repair. Of these, 59.4% (60 patients) met our criteria for the need for surgery.

Discussion

Great progress has been made in the last several years toward a better understanding of many aspects of the cleft lip and palate deformity, yet there is still a long way to go before there is agreement on the optimal treatment procedures and their timing. Internal audits are important steps in that direction for every institution or CLP team. Under this premise, the present study is an attempt to determine the frequency of need for orthognathic surgery among patients with a history of CUCLP and CBCLP at our institution.

Their phenotypical polymorphism and variable severity make incomplete clefts of the lip and palate impossible to standardize. For this reason we included in the samples only patients with complete UCLP and BCLP.

The frequency of use of orthognathic surgery carries a certain degree of bias for an institution because it is generally considered to be an unofficial indicator of the success or failure of the institution's CLP treatment protocol in terms of interference with maxillary growth. The fact that entire national restructuring efforts in cleft care (Williams et al., 2001) were partially based on Goslon Yardstick scores (an index of interocclusal sagittal relationship of plaster casts introduced by Mars et al. [1987] for patients with CLP, the last two categories of which are considered predictive of future need for orthognathic surgery), demonstrates the sensitive nature of this parameter. Consequently, very few reports have been published from institutions worldwide on frequency of use of orthognathic surgery.

The study showed that roughly 65% of patients with CBCLP and 48% of patients with CUCLP who received their primary lip and palate repairs at SickKids could benefit from orthognathic surgery. When comparing these percentages with those of patients who received primary repairs outside of our center (but at some point or another came to our facility for treatment), it is evident that a somewhat higher percentage of the non-SickKids sample seemed to require surgery (Figs. 1 and 2).

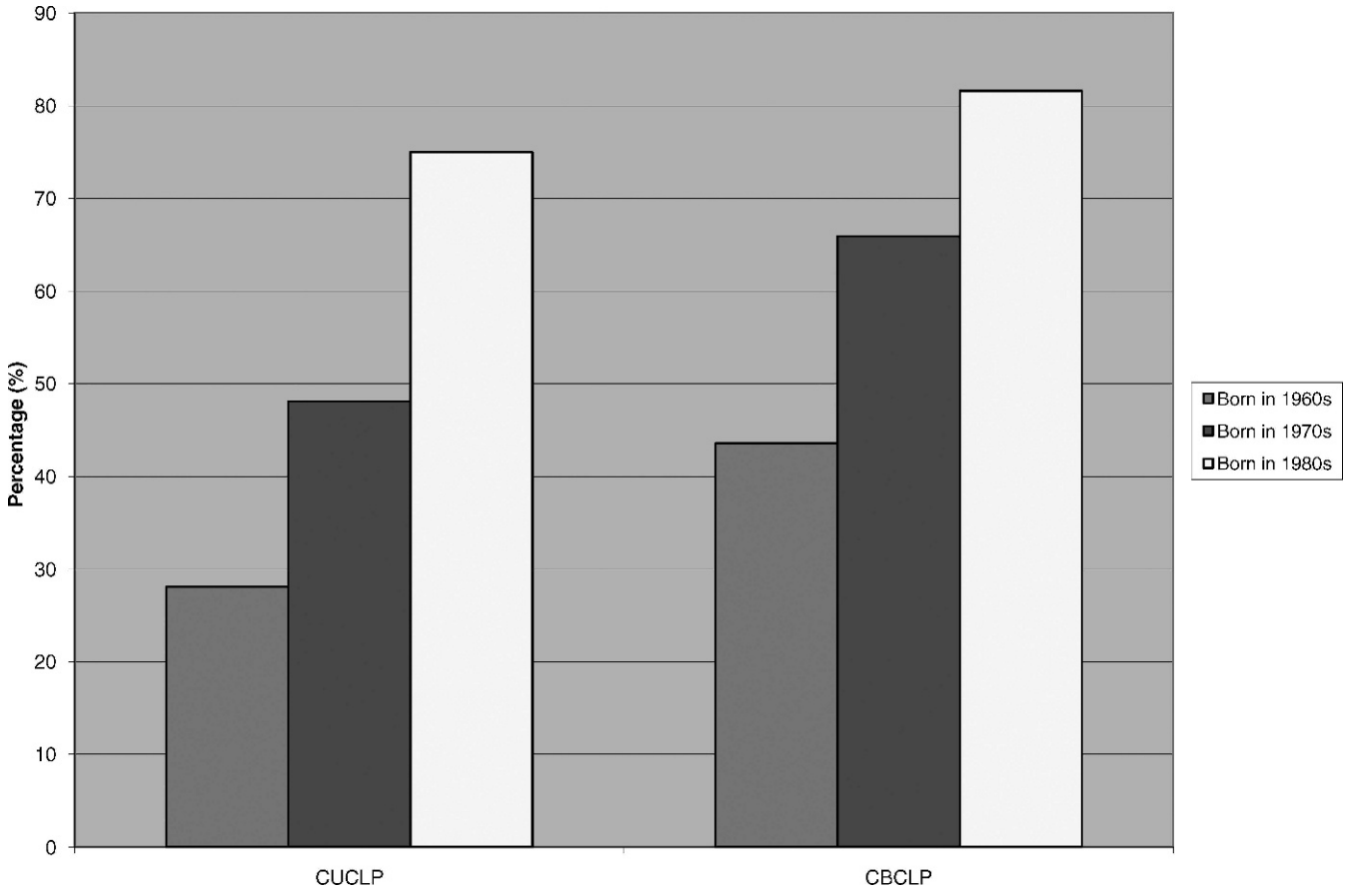

FIGURE 2 Percentages of SickKids patients deemed to require orthognathic surgery by birth decade.

Although these numbers are higher than most reported in the past, they are within the range estimated by Posnick (1991), who suggested that the need for surgery could range from 25% to 75%, depending on the criteria applied. It is interesting that these percentages are still lower than those reported recently by Good et al. (2007), which involved 76.5% of patients with CBCLP and 46.5% of patients with CUCLP. The latter report, however, is based on significantly smaller samples (17 patients with CBCLP and 33 patients with CUCLP).

Patients with a history of CLP generally present with a variable degree of midface deficiency and reduced upper lip support, which becomes more marked during their adolescence (Ross, 1987). Extraction of lower premolars and orthodontic compensation of the skeletal malrelationship could help avoid orthognathic surgery but are rarely indicated for optimal profile aesthetics in such patients. For this reason, orthodontic camouflage is seldom, if ever, attempted at our institution. We believe this to be a significant factor in explaining the relatively high orthognathic surgery rates reported.

There are, however, other variables in addition to skeletal relationship and profile aesthetics that could influence the decision to recommend/plan for orthognathic surgery: A very important such variable is cost. In Canada, the national health care plan covers the cost of the surgical procedure in its entirety, making it significantly more attractive to the average patient. A consequence of the extensive number of such procedures performed annually is raised public awareness and familiarity with the concept of orthognathic surgery, increasing the chances of acceptance of a recommended surgical plan.

Another potential reason to use orthognathic surgery for an individual with UCLP or BCLP at our institution is the presence of an ungrafted alveolar cleft in an adolescent or adult. In patients who have not had the benefit of mixed dentition bone grafting (prior to the eruption of the cleftside canine), a segmental Le Fort I osteotomy is performed to approximate the alveolar segments while grafting the residual osseous gap with iliac bone (Posnick and Tompson, 1992, 1993; Posnick, 1996). Up until the late 1990s, the protocol called for no secondary bone grafting for patients who seemed to be future orthognathic candidates. The grafting was postponed until the time of the segmental Le Fort I and performed in conjunction with that, as described above. The rationale was that if both procedures could be combined later, this would reduce the burden of care for those who evidently would require orthognathic surgery later. This protocol was changed in the year 2000 because the success of bone grafting was shown to be the highest if

performed during the mixed-dentition stage. The methods used in this retrospective study did not allow us to distinguish between patients who underwent orthognathic surgery to obliterate an ungrafted alveolar cleft from those who required correction of a sagittal skeletal discrepancy.

The results of study also showed an increasing trend for surgery by the decade of birth, which was difficult to explain. Detailed examination of the panoramic radiographs and orthodontic clinic charts revealed that a number of the patients born in the 1960s and 1970s were finished with removable prostheses and ungrafted alveolar clefts. In such patients, cleft dental gaps were expanded orthodontically to the size of two or three teeth, in order to procline the incisors enough to provide a positive overjet. Our arbitrarily set, yet somewhat rigid, cephalometric criteria were likely not sensitive enough to detect such cases, essentially underestimating the need for orthognathic surgery in patients born in the 1960s and 1970s.

The use of presurgical orthopedics was a variable briefly looked into. There was definite information available for 101 patients from the CUCLP sample. The need for surgery was slightly higher in this subsample when compared with the general CUCLP population (59.4% versus 48.3%). The procedure used in our institution for the samples assessed in this investigation was essentially a modification of the McNeil method, involving the use of a loosely fitting acrylic appliance (either passive or active) attempting to mold the alveolar segments up until the time of the lip repair (3 months). No active strapping or nasal cartilage molding was ever attempted. A recent randomized controlled study by Prahl et al. (2006) concluded that infant orthopedics with an appliance maintained up until the repair of the palate (approximately 1 year) had no effect on facial appearance of patients with a history of UCLP.

Conclusions

The goal of this study was to report objectively determined need for orthognathic surgery for patients treated at our institution. Roughly 48% of patients with CUCLP and 65% of patients with CBCLP were deemed to require orthognathic surgery. The complexity of the condition and the extent of the variability make it impossible to draw direct inferences on reasons for these seemingly elevated percentages. Our attempt to improve the profile convexity by avoiding orthodontic compensation; the Canadian Health System covering the costs of the surgery; and the inability to segregate Le Fort I advancements due to a sagittal skeletal discrepancy from those done for obliteration of an ungrafted cleft are some of the factors that we have identified as important in explaining our findings.

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