INTRODUCTION: Mandibular micrognathia is the primary defining characteristic of Pierre Robin Sequence (PRS) that sets off a physiologic cascade with varying severity of airway obstruction. The diagnosis of micrognathia is predominantly clinical, though with the advent of three-dimensional computed tomography (3D-CT), the development of objective and accurate measurements of skeletal morphology could be a useful clinical adjunct for management. The aim of this study is to investigate the efficacy of using cephalometric analysis to guide clinical management: non-operative treatment, mandibular distraction, tracheostomy.

METHODS: Records were retrospectively reviewed from 2004–2016 (IRB#2011-0247). Infants less than one year of age with PRS, evaluated for surgical management of micrognathia with a CT scan were included. 3D-CT analysis of total mandibular length (Co-Gn) and total midface length (Co-A) was performed in addition to traditional cephalometric analysis – Co-Gn, Co-A, SNA, SNB, ANB – in reformatted sagittal CT scans. Clinical data collected included age at CT scan, sex, and associated syndromic status. Chi-squared and Kruskal-Wallis tests were used to compare values among patients that were managed non-operatively, with mandibular distraction osteogenesis (MDO), and tracheostomy at any point, with Mann-Whitney U test reserved for comparing two groups.

RESULTS: 147 patients met inclusion; 33 non-operative, 73 MDO, 41 tracheostomy. CT scans were performed at an older age in the tracheostomy group compared to the non-operative and MDO groups (96.3, 58.3, 39.4 days, respectively; p=0.02). Likewise, the tracheostomy group had a greater proportion of syndromic patients compared to non-operative and MDO groups (76%, 36%, 40%, respectively; p=0.0003). Traditional cephalometric measures demonstrated no differences among the groups: Co-Gn (non-operative: 43.0mm, MDO: 41.2mm, tracheostomy: 41.1mm; p=0.17), Co-A (41.7, 40.7, 41.5mm, respectively; p=0.28), ANB (18.1°, 19.2°, 20.3°, respectively; p=0.37), SNA (84.3°, 83.0°, 84.0°, respectively; p=0.46), and SNB (66.1°, 63.8°, 63.6°, respectively; p=0.14). 3D-CT analysis of total mandibular length (46.3, 46.2, 45.2mm, respectively; p=0.48) and total midface length (49.3, 48.5, 48.5mm, respectively; p=0.37) did not differ among groups.

CONCLUSION: Cephalometric measurements from computed tomographic scans did not differ among patients that were managed non-operatively, underwent mandibular distraction, or tracheostomy. These results illustrate the limitation of solely using skeletal data as a means to predict the need for surgical intervention for airway compromise.

Reference Citations:
1. Pruzansky S, Richmond JB. Growth of Mandible in Infants with Micrognathia: Clinical Implications. Archives of Pediatrics & Adolescent Medicine. 1954;88(1):29. doi:10.1001/archpedi.1954.02050100031005.
2. Breugem CC, Evans KN, Poets CF, et al. Best Practices for the Diagnosis and Evaluation of Infants With Robin Sequence. JAMA Pediatrics. 2016;170(9):894. doi:10.1001/jamapediatrics.2016.0796.

Cost Analysis of Distraction Osteogenesis Versus Conventional Surgery in Le Fort III Surgery for Syndromic Craniosynostosis

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INTRODUCTION: Midface hypoplasia can be treated using a Le Fort III osteotomy, either via conventional surgery with immediate advancement or distraction osteogenesis to gradually advance the midface. Economic analyses are reported in as few as 0.6 percent of outcome studies in plastic surgery. There is currently no cost-effectiveness data comparing these two modalities in the literature. This study analyzes cost differences between Le Fort III conventional surgery and distraction osteogenesis among pediatric patients with syndromic craniosynostosis.
METHODS: Hospital cost-accounting databases were queried for patients undergoing single-stage advancement or distraction osteogenesis from 2007 to 2016. Nominal cost data was adjusted using the Bank of Canada Consumer Price Index. Reported costs represented the full length of stay for all utilization per patient except for anesthesia and surgeon costs (which were equivalent between groups). Parametric and non-parametric tests were used to analyze data.

RESULTS: Total costs for single-stage (n=8) were higher than distraction (n=6) (mean $57,825 versus $38,268, p<0.05). ICU costs for single-stage were significantly higher than distraction (mean, $17,746 versus $5,585, p <0.005). Distraction cases had higher OR costs than single stage but the difference was not significant (mean, $12,540 versus $9,696). Length of stay was significantly longer for single-stage patients (mean, 11 days versus 7 days, p<0.05).

CONCLUSION: This single-institution retrospective economic analysis indicates conventional Le Fort III is more costly than distraction osteogenesis. Despite higher operating room costs, recovery time led to this cost discrepancy, which is consistent with the theoretical benefits of gradual bony movements being less traumatic. The cost effectiveness ratio adds to comparative analysis of quality outcomes in existing literature and suggests distraction may provide equal clinical outcome for lower cost.

Incidence of Secondary Midface Advancement at the Time of Skeletal Maturity in Patients with a History of Early Le Fort III Distraction Osteogenesis

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INTRODUCTION: Le Fort III distraction osteogenesis is commonly recommended for children with syndromic craniosynostosis to reduce exorbitism, improve airway function, and decrease dysmorphism. This purpose of this study is to report on the long-term clinical outcomes of patients with syndromic craniosynostosis who have undergone early primary subcranial Le Fort III distraction osteogenesis and who have been followed longitudinally through skeletal maturity.

METHODS: Retrospective review of all patients who underwent Le Fort III distraction osteogenesis between the ages of 3 to 11 years and were followed throughout development with longitudinal dental, medical, radiographic, and photographic evaluations conducted through skeletal maturity and beyond. Inclusion criteria entailed having preoperative medical photographs and cephalometric studies at 6 months and 1, 5, and 10 years postoperatively after the primary Le Fort III distraction osteogenesis as well as cephalometric documentation 6 months and 1 year after the secondary midface advancement after skeletal maturity.

RESULTS: 17 patients fulfilled inclusion criteria with a mean age of 5.7 years at the time of initial Le Fort III distraction. The mean advancement of Point A was 14.9mm anteriorly and 2.7 mm inferiorly along the x- and y-axis, respectively. Orbitale moved 10.5mm anteriorly and 2.2mm inferiorly along the x- and y-axis, respectively. At 10 years postoperatively Point A moved 3.4mm anterior along the x-axis and 4.7mm inferiorly along the y-axis, while orbitale moved 0.4mm posteriorly and 3 mm inferiorly along the x- and y-axis, respectively. At the time skeletal maturity there was a return of occlusal disharmony from normal mandibular growth and a return of proptosis due to remodeling of orbitale inferiorly, and the lateral orbital rim posteriorly, while the globe continued to grow in the anterior vector. All but one study patient underwent or is scheduled to undergo a secondary midface advancement at the Le Fort III and Le Fort I level after skeletal maturity was attained.

CONCLUSION: The data demonstrates that patients who undergo early Le Fort III distraction osteogenesis before the age of mixed dentition will still most likely need a secondary midface advancement after skeletal maturity is reached given that there is a small degree of anterior growth at the level of the maxilla and no anterior growth at orbitale over time.