3-D Morphologic Characterization to Optimize Mandibular Distraction in Patients with Pierre Robin Sequence

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BACKGROUND

• Pierre Robin Sequence (PRS) triad: micrognathia, airway obstruction, glossoptosis.

• In the management of airway obstruction in patients with PRS, optimal outcomes are achieved when mandibular distraction osteogenesis (MDO) is performed as the primary surgical intervention

• At present, quantitative metrics of mandibular morphology cannot be easily translated to guide surgical planning and distractor vector selection.

PURPOSE

The aim of this study is to compare the mandibles of infants with non-syndromic PRS to controls to characterize their morphological variance in a manner relevant to distraction. This study also examines whether morphologic measurements can predict which patients will require MDO and shows how airway view grades vary among patients requiring MDO.

METHODS

• From October 2010 to March 2019, patients under 2-months-old with non-syndromic PRS were identified and age and sex-matched to control patients.

• Demographic and peri-operative data were recorded, including Cormack-Lehane airway view grades.

• Computed tomography (CT) scans from these patients were used to generate 3-dimensional (3-D) mandibular models.

• The following anthropometric landmarks were independently identified by 2 reviewers: bilateral condylions, bilateral gonions, and the menton.

• A high degree of inter-rater reliability was observed between reviewers.

• Linear and angular measurements were made. Wilcoxon rank sum and two sample t tests were performed. A *p*-value of < 0.05 was considered statistically significant.

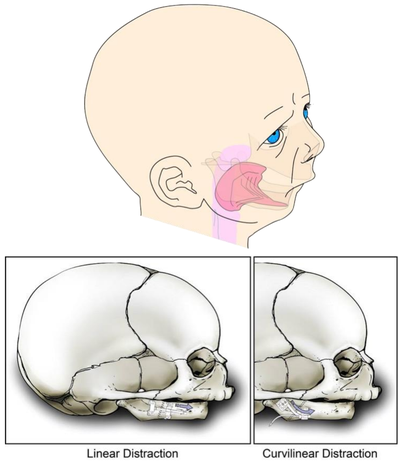

Fig 1: PRS triad and MDO vector options.

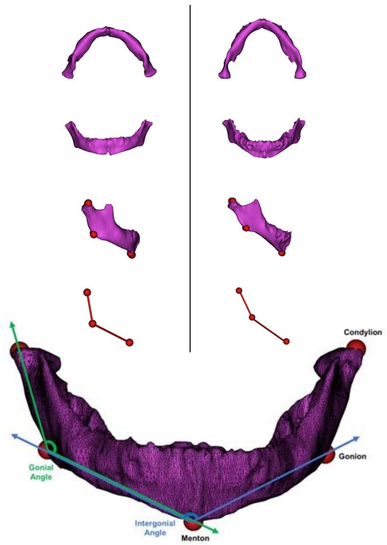

Fig 2: 3-Dimensional rendering of a mandible showing the anthropometric landmarks and angular measurements utilized.

Descriptive and Morphologic Comparison Statistics

Demographics	Pierre-Robin patients	Control patients	p
Total patients (n)	24	24	1.000
Mean age at CT, months	0.64	0.74	0.1140
Linear Measurements, mm (SD)			
Mean inter-condylar distance	57.4 (3.5)	57.7 (3.6)	0.9772
Mean inter-gonial distance	52.6 (3.9)	51.3 (4.0)	0.1793
Mean mandibular length	46.9 (4.1)	52.5 (3.5)	0.0001
Mean ramus height	16.7 (1.9)	17.3 (1.6)	0.3458
Mean mandibular body length	35.3 (3.3)	39.3 (3.2)	0.0008
Ratio (SD)			
Ramus-to-body ratio	0.47 (0.1)	0.45 (0.1)	0.0321
3-Dimensional Measurements, mm			
Inter-gonial distance	83.1 (9.1)	93.3 (7.3)	0.0007
Mandibular arc length	123.8 (11.9)	136.5 (9.2)	0.0012
Angular Measurements, degrees			
Gonial angle	125.3 (3.3)	131.3 (4.1)	<0.0001
Inter-gonial angle	94.2 (6.4)	80.4 (4.6)	<0.0001

Analysis of patients with Pierre Robin sequence based on intervention required

	MDO	Conservative management	p
Total patients (n)	17	5	
Median age at intervention, months	0.89	N/A	
Linear Measurements, mm (SD)			
Mean inter-condylar distance	57.6 (3.5)	58.4 (3.0)	0.6106
Mean inter-gonial distance	53.2 (3.8)	52.8 (3.2)	0.7244
Mean mandibular length	47.1 (4.0)	48.1 (4.8)	0.9064
Mean ramus height	16.8 (2.0)	17.2 (1.2)	0.7839
Mean mandibular body length	35.4 (3.2)	35.7 (4.1)	0.7244
Ratio (SD)			
Ramus-to-body ratio	0.5 (0.1)	0.5 (0.0)	0.8447
3-Dimensional Measurements, mm			
Inter-gonial distance	83.2 (8.8)	84.5 (10.0)	0.9064
Mandibular arc length	124.7 (3.6)	126.7 (12.6)	0.7244
Angular Measurements, degrees			
Gonial angle	124.7 (3.6)	127.1(2.2)	0.2246
Inter-gonial angle	95.1 (6.0)	93.6 (8.3)	0.5568

Mean differences of airway grades of patients requiring MDO

	Grade 1-2	Grade 3-4	p
Total patients (n)	5	9	
Linear Measurements, mm (SD)			
Mean inter-condylar distance	57.7 (1.4)	58.0 (3.2)	0.8143
Mean inter-gonial distance	53.4 (1.0)	53.5 (3.7)	0.9740
Mean mandibular length	48.6 (1.4)	47.2 (3.3)	0.3570
Mean ramus height	16.5 (2.3)	17.2 (1.1)	0.4514
Mean mandibular body length	37.1 (1.9)	35.2 (2.8)	0.1890
Ratio (SD)			
Ramus-to-body ratio	0.4 (0.1)	0.5 (0.0)	0.1818
3-Dimensional Measurements, mm			
Inter-gonial distance	88.4 (4.0)	81.7 (8.5)	0.1246
Mandibular arc length	128.8 (5.9)	123.8 (10.8)	0.3618
Angular Measurements, degrees			
Gonial angle	125.7 (3.2)	124.7 (4.3)	0.6434
Inter-gonial angle	91.4 (5.4)	95.8 (6.1)	0.1962

RESULTS

- A total of 24 patients (mean age 0.64 months) with a diagnosis of non-syndromic PRS and 24 control patients were included.
- Patients with PRS were found to have shorter ramus heights (16.7mm vs 17.3mm; p=0.346) and shorter mandibular body lengths (35.3mm vs 39.3mm; p<0.001) compared to controls. Gonial angles were more acute (125.3° vs 131.3°; p<0.001) and inter-gonial angles were more obtuse (94.2° vs 80.4°; p<0.001) in PRS patients.
- Of the PRS patients, 17 required MDO, 5 were managed conservatively, and 2 required other management methods.
- No significant differences in mandibular measurements were found among patients requiring MDO vs conservative management, nor among patients requiring MDO with high vs low airway grades.

CONCLUSION

• Our study examines both the largest and youngest PRS patient population to date, making it the most relevant regarding management of early airway obstruction with MDO.

• The observed statistically significant differences in mandibular body length and inter-gonial angle suggest that univector distraction of the mandibular body should allow for normalization of mandibular morphology in patients with non-syndromic PRS.

• Our findings also indicate that determining the optimal treatment modality for airway obstruction in patients with PRS remains a clinical decision.

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