Functional outcomes of mandibular distraction for the relief of severe airway obstruction and feeding difficulties in neonates with Pierre Robin sequence

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SUMMARY: Konaş E, Çalış M, Bitik O, Yiğit Ş, Korkmaz A, Yurdakök M, Tunçbilek G. Functional outcomes of mandibular distraction for the relief of severe airway obstruction and feeding difficulties in neonates with Pierre Robin sequence. Turk J Pediatr 2016; 58: 159-167.

The purpose of this study was to review the application of mandibular distraction to relieve severe airway obstruction or feeding problems of neonates. Thirteen neonates with Pierre Robin sequence who underwent bilateral mandibular distraction between 2010 and 2013 for relief of their severe airway obstruction or feeding problems were retrospectively reviewed. The mean preoperative and postoperative airway diameters were 3.89±1.64 and 9.03±1.98 mm. respectively and significant difference was observed with distraction ($p<0.001$). The rate of severe airway infection also significantly decreased from 69.2% to 23.1% ($p=0.016$). 84.6% of the patients were able to be fed orally at discharge whereas 6 patients (46.2%) required support via orogastric tube before distraction ($p=0.125$). No growth disturbance, dental complications or malocclusion was observed in the long-term follow up. Mandibular distraction appears to be a promising and effective surgical option for relieving airway obstruction and feeding problems in severe Pierre Robin Sequence patients.

***Key words:** airway obstruction, feeding problems, neonates, Pierre Robin Sequence, mandibular distraction.*

Pierre Robin sequence (PRS) is characterized by the classic description of the pathology with presence of micrognathia, glossoptosis and cleft palate. The major physiological restrictions of these children are breathing and feeding problems secondary to abnormally small jaw leading to obstruction by tongue[1].

The spectrum of modalities to relieve airway problems of PRS patients vary from supportive measures to surgical interventions. As this airway obstruction may be compensated as the craniofacial skeleton grows, the majority of the neonates respond to supportive interventions such as prone positioning and continuous positive airway pressure. In despite of non-invasive modalities, up to 23% of the children may require hospitalization and invasive interventions such as intubation and tracheostomy[2].

For many years tracheotomy was accepted as the final option for neonates with severe airway obstruction. Although tracheotomy is a lifesaving procedure, it lengthens hospitalization, increases health care costs[3] and is associated with secondary complications related to lack of proper homecare[4].

Following the initial applications of McCarthy in the early 1990's, distraction osteogenesis became one of the most popular surgical intervention for the craniofacial skeleton. Later on, mandibular distraction osteogenesis (MDO) appeared to be an alternative to eliminate the need for tracheotomy in PRS patients with severe airway obstruction[5].

In this article we report our experience on mandibular distraction osteogenesis for neonates with PRS. The purpose of this singlecenter retrospective study is to review the application of mandibular distraction to relieve severe airway obstruction or feeding problems of these patients and to discuss the advantages and possible pitfalls.

Material and Methods

Patients:

This study retrospectively reviewed 13 neonates with Pierre Robin Sequence who underwent bilateral mandibular distraction between 2010 and 2013 following the approval of Institutional Review Board (Approval No: GO 14/153). Neonates were referred from the neonatology unit and were followed there at the postoperative period. Neonates with clinical features of Pierre Robin sequence (Fig. 1) and respiratory distress related to mandibular deficiency (micrognathia and glossoptosis), those with a follow up of at least one year, having radiographic evaluation of both preoperative and postoperative period are included in the study. Cleft palate was not a required feature.

Standard demographic data of patients that included sex, birth weight, gestational age, prematurity, associated malformations and comorbidities were recorded. The perioperative variables of the patients such as age of distraction placement, initial maxillomandibular discrepancy (MMD) (Fig. 2), feeding history, total length of distraction, total length at the time of extubation, postoperative day of extubation, weight at operation and at certain intervals during follow up, postoperative complications, preoperative infection and feeding status and status at discharge were recorded.

Surgical Method and Distraction Protocol: The operations were performed under general anesthesia following preoperative evaluation of the mandibular anatomy by anteroposterior and

Table I. Summary of the Demographic Data of Patients

Patient	Gestational age	Birth weight (g)	Sex	Cleft palate	Other malformations and associations
1	37w 3d	2,800	Female	+	-
2	40w 0d	2,400	Female	-	Cleft no 7, interhemispheric cyst, atrial septal defect
3	35w 1d	2,620	Male	+	-
4	38w 3d	2,250	Female	+	Partial trisomy 7q, partial monosomy 8p
5	39w 1d	3,050	Male	+	Pectus excavatum
6	38w 2d	2,850	Male	+	Patent ductus arteriosus
7	39w 2d	3,200	Female	+	-
8	39w 0d	3,260	Female	+	Atrial septal defect
9	38w 0d	2,980	Female	+	Atrial septal defect
10	40w 0d	3,200	Male	+	Minor auricular deformity (accessory crus)
11	39w 4d	3,200	Male	-	-
12	40w 3d	3,410	Female	-	Ventricular septum hypertrophy, patent foramen ovale
13	38 w 0d	2,900	Male	+	-
N=13		2,932.31±346.22*	Male 6 (46.15%)***	Yes 10 (76.92%)***	
		2,980 (2,250-3,410)**	Female 7 (53.85%)***	No 3 (23.08%)***	

*Mean±SD; **median (minimum-maximum); *** n (%)

Table II. Summary of the Airway Management and Perioperative Data Related with Distraction

Patient	Preoperative airway management	MMD (mm)	Total length of distraction (mm.)	Preop. airway diameter (mm.)	Postop. airway diameter (mm.)	Length of distraction at extubation (mm)	Postop. day of extubation	Preop. infection	Postop. infection
1	INT	14	16	3.1	8.7	6	6	Pneumonia	-
2	POS	20	20	6.7	8.9	3	4	Pneumonia	-
3	POS	13	15	2	7	1,5	1	-	-
4	POS	18	19	4,7	12.6	0	0	Sepsis	Bronchitis
5	POS	12	13	5.4	9.2	0	0	-	-
6	POS	13	14	6.4	10.2	0	0	-	-
7	INT	13	14	2.6	5.3	9	8	Pneumonia	Pneumonia
8	CPAP	13	14	3.6	9.7	3	4	-	-
9	POS	12	13	4.2	7.4	6	6	Bronchitis	-
10	POS	14	14	3.9	9.4	0	0	Sepsis	Pneumonia
11	INT	14	16	4.2	11.6	3	4	Sepsis	-
12	CPAP	15	16	1.1	7.2	3	4	Pneumonia	-
13	INT	14	16	2.7	10.2	9	6	Bronchitis	-
N=13	POS 7(53.8)*** INT 4(30.8)*** CPAP 2(15.4%)***	14.23±2.31* 14 (12-20)**	15.38±2.14* 15 (13-20)**	3.89±1.64* 3.90 (1.10-6.70)*	9.03±1.98* 9.20 (5.3-12.6)*	3.35±3.25* 3 (0-9)**	3.31±2.81* 4 (0-8)**	NO 4 (30.8%)*** YES 9 (69.2%)***	NO 10 (76.9%)*** YES 3 (23.1%)***
				P<0.001				P =0.016	

INT: endotracheal intubation; POS: positioning; CPAP: continuous positive airway pressure mask
Paired t-test – Mc Nemar test; *mean±SD; **median (minimum-maksimum); *** n (%)
PAIRED T TEST - MC-NEMAR TEST *Mean ± Std. **Median (Maximum-Minimum) ***n(%)
MMD: Maxillomandibular discrepancy

Table III. Summary of Feeding History and Weight Gain

Patient	Weight at operation (g.)	Weight at discharge (g.)	Postoperative weight at 1 mo. (g.)	Postoperative weight at 3 mo.(g.)	Postoperative weight at 6 mo.(g.)	Preoperative Feeding	Feeding at Discharge
1	2,450	2,900	3,700	5,650	8,000	PO	PO
2	6,500	7,450	6,950	7,500	8,500	OG	PO
3	3,370	4,100	4,800	6,800	8,100	PO	PO
4	2,640	3,000	4,000	5,000	5,500	PO	PO
5	8,100	8,300	8,700	9,900	12,300	OG	OG
6	5,000	5,000	7,000	7,400	9,000	PO	PO
7	2,930	3,100	3,300	3,500	4,800	OG	PO
8	3,220	3,630	4,400	6,000	7,700	PO	PO
9	3,010	3,400	3,880	5,500	7,100	PO	PO
10	4,500	4,650	4,800	5,300	6,820	PO	PO
11	3,900	3,800	4,100	4,900	6,100	OG	PO
12	3,800	3,200	3,300	5,000	7,000	OG	OG
13	2,710	3,180	3,250	3,900	5,500	OG	PO
N=13	4,010.00±1,668.46*	4,285.38±1,723.26*	4,783.08±1,705.82*	5,873.08±1,697.62*	7,416.92±1,935.90*	OG 6 (46.2%)***	OG 2 (15.4%)***
	3,370 (2,450-8,100)**	3,630 (2,900-8,300)**	4,100 (3,250-8,700)**	5,500 (3,500-9,900)**	7,100 (4,800-12,300)**	PO 7 (53.8%)***	PO 11 (84.6%)***
						P = 0.125	

McNemar Test; *mean ± SD; **median (minimum-maximum) ; ***n(%); OG: orogastric; PO: peroral

Fig. 1. Preoperative appearance of a patient with micrognathia and severe respiratory distress

Fig. 2. Maxillomandibular discrepancy is measured by placing a ruler in the midline against the mandibular alveolus. There was 17 mm. of disharmony measured for this patient.

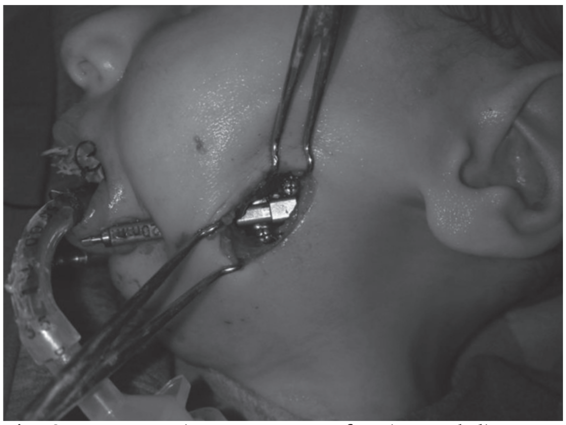

Fig. 3. Intraoperative appearance of an internal distractor with 20 mm. of length following bicortical mandibular osteotomies.

lateral radiograms. The bilateral mandibular distraction was performed by percutaneous approach using inframandibular incision. Bone was exposed by subperiosteal dissection following blunt dissection of the overlying soft tissue and stripping masseteric muscle fibers. Internal distractors (Synthes Inc, Oberdorf, Switzerland) with either 20 or 25 mm. of length (Fig. 3) were placed at the first step, and complete bicortical inverted L shaped mandibular osteotomies were performed preserving both the inferior alveolar nerves and the tooth buds. Proximal and distal footplates of the distractors were fixed using screws with 1.0 mm. of diameter (Synthes Inc, Oberdorf, Switzerland). Extension rods were used in all patients. Distractors were activated intraoperatively to ensure the complete osteotomy. Distraction was initiated following 48 hrs. of latency at a rate of 1.5 mm/day (twice daily with the rhythm of 0.75 mm. per each) and continued until a slight overcorrection was reached. Distractors were removed after 12 weeks of consolidation (Fig. 4).

Evaluation of the Airway: The distance between posterior pharyngeal wall and base of tongue was measured to calculate the cross-sectional area of the airway using the preoperative and postoperative standardized lateral head radiographs (Fig. 5A-5B). Postoperative evaluation was made at the end of the consolidation period, at the time of distractor removal in all patients. In order to evaluate the efficacy of distraction, besides comparison of preoperative and postoperative airway areas, bone distraction was also compared with its reflection on airway distance.

Statistical Analysis

Statistical analysis was performed using the SPSS 21.0 for Windows (Statistical Package for Social Sciences; SPSS Inc, Chicago, IL). Shapiro-Wilk test was used to evaluate the normal distribution pattern of the quantitative data. Descriptive statistical values were expressed as mean ± standard deviation (Std) and median (maximum-minimum). Paired Sample T test was used to compare the preoperative and postoperative airway diameters. Infection and feeding status were analyzed using McNemar (exact) test. P values less than 0.05 were considered as statistically significant.

Results

Demographic information of 13 neonates included in the study is summarized in Table I. Among those, 7 patients (54%) were female and 6 patients (46%) were male. Only one

Fig. 4. Appearance of the patient at the end of consolidation period. Note the weight gain and relief of the respiratory distress. The scar is almost invisible.

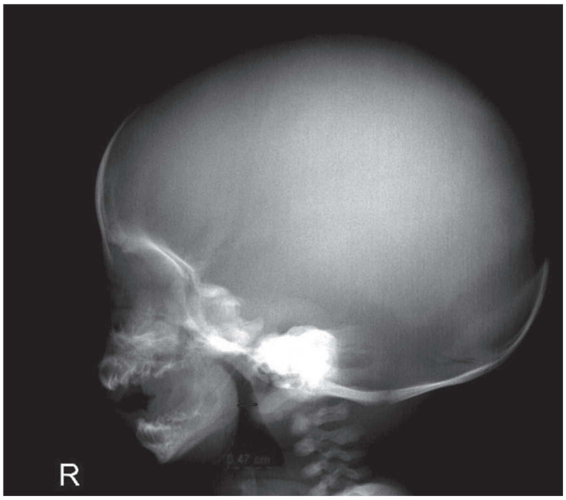

Fig. 5a. Evaluation of the preoperative lateral head radiograph of patient 4 reveals airway diameter of 4.7 mm.

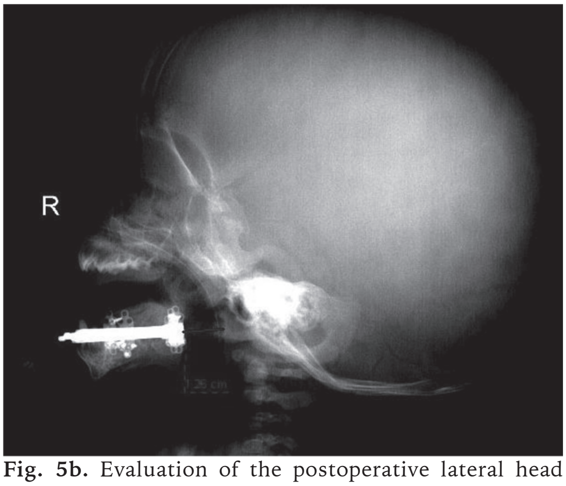

Fig. 5b. Evaluation of the postoperative lateral head radiograph of patient 4 reveals airway diameter of 12.6 mm.

patient was premature (<37 weeks). Mean birth weight of patients was 2,932.31±346.22 g and one patient had low birth weight (<2,500 g). Ten of the patients with PRS (76.92%) also had cleft palate.

Airway management and perioperative information related to distraction were summarized in Table II. Even though 7 patients (53.8%) were managed by positioning, 2 patients (%15.4) were followed by supplemental oxygen via continuous positive airway pressure (CPAP) mask and 4 patients (30.8%) required endotracheal intubation. Mean MMD of the patients were 14.23±2.31 mm. and the mean length of distraction planned for correction was 15.38 ± 2.14 mm. The mean preoperative and postoperative airway diameters were 3.89 ± 1.64 and 9.03 ± 1.98 mm. respectively and significant difference was observed by mandibular distraction ($p<0.001$). The rate of severe airway infection also significantly decreased from 69.2% to 23.1% ($p=0.016$).

The feeding history and weights of the patients at various intervals are summarized in Table III. The mean body weight has increased from 4,010.00 ± 1,668.46 g to 4,285.38 ± 1,723.26 g between the interval from the operation to discharge. With respect to nutrition, 6 patients (46.2%) required support via orogastric tube before distraction, whereas 84.6% of the patients were able to be fed orally at discharge ($p=0.125$).

There were no major surgical complications except nosocomial pneumonia treated by systemic antibiotics in two patients. Besides minor complications such as extension rod fracture observed in one patient and pin tract infection treated in 2 patients, unilateral injury of marginal mandibular branch of facial nerve was experienced in one patient. One patient required re-intubation secondary to development of respiratory distress following extubation in the neonatology unit. The patients healed with cosmetically and functionally acceptable clinical results. No growth disturbance, dental complications or malocclusion were observed in the long-term follow up (Fig. 6A-6B).

Discussion

Respiratory distress due to backwards lowering of the tongue (glossoptosis) in micrognathic patients were initially reported by Pierre Robin[6],

Fig. 6a. Anterior view of the patient three years after distraction.

Fig. 6b. Lateral view of the patient three years after distraction.

a French otolaryngologist, in 1923. Many terms have been appended for this clinical presentation such as "syndrome"[7], "anomalad"[8] and recently sequence[9]. Pierre Robin sequence is currently defined with the classic triad of micrognathia, glossoptosis and in approximately one third of patients, cleft palate[10].

PRS is a rare congenital anomaly with average incidence of 1:8,500[1]. Clinical presentation of PRS may exist as an isolated anomaly or may be part of additional anomalies of a syndrome[11]. The leading hypotheses related to PRS are compression of the mandible secondary to intrauterine position of the fetus, pharmacologic teratogenicity, genetic mutation and sporadic etiology[11,12]. PRS cases with severe obstruction leads to feeding problems, dehydration, exhaustion, failure to thrive and in extended cases cardiac problems and death[2].

Nonsurgical modalities such as positioning and feeding strategies are sufficient to provide support for the majority of cases in respect to the fact that airway obstruction may be compensated as the craniofacial skeleton grows[13]. For severe cases besides proper positioning, nonsurgical interventions range from supplemental oxygen support via mask to intubation[11]. Leading surgical interventions to prevent glossoptosis are tongue-lip adhesion[1] and release of suprahyoid muscles[14]. In extreme cases to provide long-term support and relieve airway obstruction tracheotomy procedure is inevitable[15].

Traction of the micrognathic mandible to correct the deformity and relieve the airway obstruction had been studied for decades. In 1937, Callister[16] reported the first surgical treatment of a PRS patient by spring traction of the mandible for weeks using a halo device integrated into a back brace. In 1949, Longmire and Sanford[17] proposed orthopedic traction by circumandibular wires attached to pulleys and weight hangers. Development of surgical treatment of micrognathia was deferred as the mentioned techniques were transmitting the traction forces directly to the joint and leading to temporomandibular joint ankylosis[16].

Mandibular advancement with sagittal split osteotomy demonstrated successful results for correction of adult obstructive sleep apnea by tongue base advancement as well as the mandible[18,19]. In the early 1990's, the initial applications of distraction osteogenesis in the craniofacial skeleton by McCarthy et al[21]. popularized mandibular distraction in plastic surgery[20]. Distraction of the mandible, besides lengthening the bony mandible, also pulls the tongue forward by anterior advancement of the related muscle and soft tissue[16]. These studies brought about the idea that mandibular distraction may also be beneficial for correction of airway obstruction.

Conventional approach relies on mandibular catch up phenomenon and delays surgery for PRS hoping for spontaneous correction of the deformity[22]. Even though an acceleration is observed in the postnatal period, in cases with severe airway obstruction, this growth spurt is insufficient to advance tongue base and relieve the persisting airway obstruction[1,23]. Besides, following the patients with positional management may lead to lengthy hospitalization periods and probable relief of the airway obstruction with tracheotomy. In their study with 6 neonates with PRS and severe airway obstruction, Denny et al[5]. used mandibular distraction to avoid tracheotomy. More recently Sesenna et al[24]. demonstrated the benefit from mandibular distraction in PRS patients for relieving airway problems and preventing tracheotomy procedure.

In our study, we reviewed our experience with 13 children followed with PRS and underwent inverted L shaped mandibular osteotomies and distraction to relieve severe airway obstruction and feeding difficulties. External approach is preferred for placing internal distractors as exposure via intraoral incisions may lead to contamination of the flora and secondary infections. Inverted L shaped osteotomy was preferred to preserve tooth buds and not to lead to any dental complications. Second precaution to keep surgical site away from the tooth buds was to place the distal footplate of the distractor close to the inferior cortical rim of the mandibular border away from the tooth buds and to use relatively thin screws with 1.0 mm. of diameter for fixation. One of the most significant findings of our study was that mandibular distraction allowed us to extubate neonates sooner and prevent them from probable tracheotomy procedure. In respect to our personal observation we could propose that at least 6 mm of distraction was necessary for safe extubation and may be accepted as a clinical cut off limit.

Besides bringing the micrognathic mandible forward in means of aesthetic appearance, it is also demonstrated that mandibular distraction significantly increases the airway diameter in quite a short interval, alleviating the symptoms related to airway obstruction. Distraction has also been observed to be beneficial in preventing airway infections and its systemic manifestations such as sepsis.

Another consequence of PRS is associated feeding problems[25]. Some neonates are not able to be fed orally and necessitates the placement of nasogastric tube[11]. Several theories have been proposed related to this restriction such as glossoptosis preventing the forward positioning of tongue during swallowing and additional lack of sucking and swallowing coordination secondary to airway obstruction in severe patients[26]. Mandibular distraction had also found to be beneficial in means of feeding problems[1,11]. In our study, in 4 patients, whom had been fed via nasogastric tube in the preoperative period, oral feeding had been initiated after distraction.

Another remarkable finding of our study is that mandibular distraction appears to be relatively safe procedure even in the early neonatal period. No major surgical complication was observed whereas two patients were treated by wide spectrum antibiotics for nosocomial pneumonia in the early postoperative period. One of the most striking surgical complications was the injury of marginal mandibular branch of facial nerve in a single patient (7.7%) in spite of blunt dissection for exposure of the bone. Even though mandibular distraction appears to be a safe procedure in the neonates, reports related to long-term effects are limited. Tibesar et al[27]. retrospectively reviewed and reported open bite deformity (28%), dental complications (16%) and facial nerve injuries (9%) as the leading complications in the long-term period.Our complication rates appear to be comparable with the mentioned previous studies.

Even though mandibular distraction is one of the most popular and well known procedures in craniofacial surgery, currently the studies related to outcomes of mandibular distraction following inverted L shaped osteotomies, in neonates with Pierre Robin Sequence to overcome severe airway obstruction and feeding restrictions are limited. In summary, our study is important to emphasize the beneficial effect of mandibular distraction in increasing the airway diameter besides obtaining an aesthetic facial profile by bringing retrognathic mandible forward and preventing airway infections and its systemic manifestations such as sepsis. Restriction of expected craniofacial growth, effect of distraction on dentition and speech in long-term should be evaluated by further studies.

Conclusion

Mandibular distraction following bicortical inverted L shaped osteotomies appears to be promising and effective surgical option for relieving airway obstruction and feeding problems in severe PRS patients. It provides stable and permanent clinical benefits and avoids morbidity associated with tracheotomy. Long-term results of on facial growth, dentition and speech should be evaluated in further studies.

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