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Review Article

Feeding and reflux in children after mandibular distraction osteogenesis for micrognathia: A systematic review

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ABSTRACT

Mandibular distraction osteogenesis (MDO) is becoming increasingly more commonly used as in neonates and infants with upper airway obstruction secondary to micrognathia. A significant number of these children are dependent on nasoenteric feeding or gastrostomies after birth for adequate nutrition and often suffer from gastro-esophageal reflux (GERD).

Objective: This analysis is a subset of a larger systematic review. The objective of this study is to determine the effects of MDO on feeding and GERD.

Data sources: The databases searched included PubMed, Embase, Scopus, Web of Knowledge and grey literature sources.

Study selection: The inclusion criterion included studies in children with clinical evidence of micrognathia/Pierre Robin Sequence (PRS) who have failed conservative treatments, including both syndromic (sMicro) and non-syndromic (iPRS) patients. 21 studies relevant to feeding and 4 studies relevant to GERD outcomes were included. All studies included were case series and case reports.

Results: MDO leads to a significant improvement in feeding, with 82% of children feeding exclusively orally after surgery. The overall percentage of children with iPRS who were feeding orally was 93.7% compared with only 72.9% in the sMicro group ($p < 0.004$). A growth decline within the first six weeks after surgery was observed in multiple studies. Overall, out of 70 patients with pre-operative GERD, only four had evidence of GERD after surgery.

Conclusions: Considering the limitations of this systematic review, this study found that successful relief of airway obstruction by MDO leads to improvement of feeding and improvement in symptoms of GERD in children with upper airway obstruction secondary to micrognathia. Clinicians need to be aware of the risk of growth decline in the initial post-operative period.

Contents

Abbreviations: MDO, mandibular distraction osteogenesis; GERD, Gastro-esophageal reflux; PRS, Pierre Robin Sequence; iPRS, isolated Pierre Robin Sequence; sMicro, syndromic micrognathia; RR, retrospective review; CR, Case report; TMJ, Temporomandibular Joint.

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1. Introduction

In 1923, the French stomatologist Pierre Robin was the first to describe a constellation of symptoms associated with upper airway obstruction in neonates now known as Pierre Robin Sequence (PRS) [1]. This sequence is a craniofacial anomaly characterized by mandibular micrognathia, glossoptosis, and in most cases a 'U' shaped cleft palate. Micrognathia is a congenital condition characterized by an abnormally small mandible. This condition tends to occur in conjunction with posterior tongue displacement (glossoptosis), which can lead to physical obstruction of the oropharyngeal and hypopharyngeal regions on inspiration. There is only limited epidemiological data, but the incidence has been reported to range from approximately 1 in 8500 live births in Merseyside [2] to 1 in 14,000 live births in Denmark [3]. The most recent study from Germany reported an incidence of approximately 1 in 8000 births [4]. This variation in incidence is related in part to the inconsistent definition of PRS in the literature.

Diagnosis of patients with PRS is challenging due to the wide spectrum of PRS phenotypes, variation in degree of airway obstruction, feeding difficulties and the need for treatment. This has led to some authors only characterizing those with airway obstruction needing treatment as having PRS [5], others will include all patients with micrognathia and glossoptosis, or limit the PRS diagnosis to those with associated cleft palates as having PRS [6]. Although these clinical features are most commonly seen in isolation [7], they can also occur in association with other clefting conditions of the craniofacial skeleton; for example, Treacher Collins Syndrome, Stickler syndrome and Nager syndrome. Such co-occurrences further complicate the diagnosis. For simplicity, in this review, those without an associated syndrome have been referred to as isolated PRS (iPRS) and those with an associated syndrome referred to as syndromic micrognathia (sMicro).

The micrognathia and upper airway obstruction contribute to a wide range of clinical problems in these children [8,9]. These infants have significant feeding and swallowing problems. During the normal swallowing process, breathing is suppressed and a decrease in ventilation occurs during the sucking process [10]. In contrast to normal infants, infants with upper airway obstruction may have to increase the efforts to breathe even at rest, and hence may lack the pulmonary reserve necessary to support the additional respiratory effort required for oral feeding. In addition to this, these children also tend to have higher caloric consumption due to their repeated attempts to clear their upper airway. All of these factors may cause failure to thrive [11,12]. These children are often dependent on long-term nasoenteric feeding or gastrostomy feeding due to these issues [13].

Another significant complication is the increased incidence of gastro-esophageal reflux (GERD) in children with PRS. It is hypothesized that upper airway obstruction results in an increased inspiratory effort to overcome the obstruction, which results in a negative intra-thoracic pressure that can cause a suction type effect on the gastric contents [14].

All of these complications are believed to be secondary to the upper airway obstruction; therefore the priority is to treat the upper airway obstruction. However, it is yet to be determined if relieving the upper airway obstruction will also resolve these complications. The purpose of this systematic review was to identify and synthesize the best available evidence on the effect of MDO on feeding and gastro-esophageal reflux in children with upper airway obstruction secondary to micrognathia. This paper reports a sub-set of the results of a larger systematic review, which also evaluated the overall effectiveness of MDO at relieving airway obstruction [15].

2. Methods

This systematic review was conducted as part of fulfillment of a Masters of Clinical Science degree at the University of Adelaide. The review was conducted according to a peer reviewed protocol prepared a priori [16]. The PICO (Participants, Intervention, Comparator, Outcomes) criteria used for this review are included in Table 1. All the studies included were children with clinical evidence of micrognathia who underwent bilateral MDO. All studies needed to have included the conservative treatment options attempted, reasons for failure, and minimum of 1 year follow up. Syndromic and non-syndromic children were included if there was clinical evidence of glossoptosis and upper airway obstruction, but some particular conditions were excluded. These include bilateral TMJ ankylosis, unilateral hemifacial microsomia or other conditions that may be contributing to the airway obstruction for reasons other than the micrognathia alone. Also, children with known lower airway abnormalities prior to treatment were also excluded.

The types of studies considered included both experimental and epidemiological study designs including randomized control trials, quasi-experimental studies, prospective and retrospective cohort and case control studies. The review also considered case series and case reports.

The search strategy included both published and unpublished studies in English from 1990 to November 2013. The databases searched included PubMed, CINAHL, EMBASE, SCOPUS, Web of knowledge and other grey literature databases such as Scirus and Mednar. The search strategy optimized for PubMed is detailed in Fig. 1. Search terms and strategy were translated for use with alternative databases.

Papers selected for retrieval were assessed by two independent reviewers for methodological validity and quality and critically

Table 1
PICO criteria for the systematic review.

Participants	- Male and female children from birth with clinical evidence of micrognathia - Clinical evidence of upper airway obstruction with failed conservative treatments - Syndromic and non-syndromic children - Bilateral mandibular distraction Exclusion - Children who underwent unilateral distraction - Children with known pre-operative central apnea/lower airway abnormalities - TMJ ankylosis/hemifacial microsomia or other mandibular condition leading to airway obstruction
Intervention	Bilateral mandibular distraction osteogenesis
Outcomes	Feeding Gastro-esophageal reflux