Mandibular distraction osteogenesis in the management of airway	y
obstruction in children with micrognathia: a systematic review	

Submitted by

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A thesis submitted in total requirements for the degree of Master of Clinical Science

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August 2015

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In the name of God, Most Compassionate, Most Merciful

To my Father, and my Father's Father......

Acknowledgements

I extend my immeasurable appreciation and deepest gratitude for the help and support extended to the following people who in one way or another have contributed to making this thesis possible.

My supervisors, Dr David Tivey, Dr Kandiah Umapathysivam and Professor Peter Anderson for their patience, advice, editing and constant motivation. Without such dedicated supervisors, this thesis would not be what it is today. I am forever grateful to have met such incredible academics and scholars.

The librarian at the University of Adelaide library, Maureen Bell, for her teaching and advice on how to properly conduct a literature search through different databases.

My father Hamdi Breik and mother Randa Buraik whose love and inspiration have been my pillars of strength since birth. Despite the challenges they have faced in life, their wisdom and love for knowledge have always been an inspiration to me. All my dreams and achievements are dedicated to them.

Dana, Lina and Nada – the most compassionate and caring sisters a person could ever ask for. Their unconditional love gives me strength every day.

Most importantly I wish to thank God for His guidance and for giving me the strength to dedicate my career to the care of His people. Thank you God for the colleagues and peers I have worked with over the years, and for the beautiful patients I have had the privilege of treating.

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Executive summary

Background

Mandibular distraction osteogenesis (MDO) is becoming increasingly commonly used as the primary surgical option for neonates and infants with upper airway obstruction secondary to micrognathia or to facilitate decannulation for tracheostomy dependent children.

Objectives

The objective of this review was to identify and synthesize the best available evidence on the effectiveness of MDO on airway patency, feeding, gastro-esophageal reflux (GORD) and long-term development in children born with upper airway obstruction secondary to micrognathia. This review also aims to determine the ideal rate of distraction, and compare outcomes of external and internal distractors in this patient group.

Inclusion criteria

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 isolated PRS (iPRS) patients. The intervention is patients who have undergone bilateral distraction osteogenesis to prevent a tracheostomy or to facilitate decannulation. The comparator intervention is patients who underwent a tracheostomy alone. The outcomes of interest include relief of airway obstruction with MDO, decannulation of tracheostomy dependent patients, feeding and reflux changes, surgical outcomes such as comparison of rate of distraction and type of distractor. All study designs were included.

Methods

The databases searched included PubMed, Embase, Scopus, Web of Knowledge and grey literature sources. Of the 4815 studies found in the initial search, only 66 were included after critical appraisal. Due to the nature of the studies included, a meta-analysis was not possible. The data was pooled by calculating weighted means.

Results

Primary MDO for the relief of upper airway obstruction was successful in 95% of cases in the literature. Syndromic (sMicro) patients had odds of failure that were four times higher than those of iPRS patients. The most common causes of failure are previously undiagnosed lower airway obstruction, central apnoea, undiagnosed neurological abnormalities and complex multiorgan anomalies. Mandibular distraction osteogenesis (MDO) was less effective (80.3% success rate) at facilitating decannulation of tracheostomy dependent children. Failure in these patients was most commonly due to severe preoperative gastro-oesophageal reflux disease (GORD), swallowing dysfunction and tracheostomy related complications. The failure rate was higher when MDO was performed at an age of ≥24 months for this group of patients. Approximately 84% of children can be exclusively oral fed after MDO. The odds of needing feeding adjuncts were five times higher in syndromic children. There was a trend towards a growth decline in the first six weeks after surgery. MDO relieves GORD in the majority of patients. Patients who were tracheostomy dependent with severe GORD were at higher risk of failure to decannulate after MDO. There was no difference in success rate when comparing a distraction rate of 1mm/day with 2mm/day. External distractors were associated with a higher rate of failure and complications compared to internal distractors. Overall, there was a paucity of long-term results in the literature. Recurrence of airway distress may occur due to a relapse of retrognathia or TMJ ankylosis.

Conclusion

Mandibular distraction osteogenesis is an effective technique for preventing tracheostomy in children with airway obstruction secondary to micrognathia (Level 4 evidence). Thorough airway evaluation and sleep study pre-MDO is necessary to exclude multilevel airway obstruction and central apnoea. Mandibular distraction osteogenesis has a slightly lower success rate at facilitating decannulation. Thorough airway evaluation, assessment for reflux and swallowing dysfunction are necessary prior to surgery. Mandibular distraction osteogenesis is effective at alleviating feeding problems and reflux symptoms in these children. Care needs to be taken to avoid a general growth decline that has been reported in the first six weeks after surgery. Distracting at a rate of 1mm/day or 2mm/day below the age of 12 months is safe. Internal distractors have a higher success rate and a lower rate of complications than external distractors. More studies are needed to evaluate the long-term implications of MDO on facial development and long-term complications.