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# Tracheostomy versus mandibular distraction osteogenesis in infants with Robin sequence: a comparative cost analysis☆

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**Abstract**

Many treatments have been described for infants with Robin sequence and severe respiratory distress, but there have not been many comparative studies of outcome and cost-effectiveness. The aim of this study was to compare the cost and complications of two common interventions – mandibular distraction osteogenesis and tracheostomy. Nine patients with isolated Robin sequence (mandibular distraction osteogenesis, $n=5$, and tracheostomy, $n=4$) were included in the analyses. Predetermined costs and complications were obtained retrospectively from medical records and by questionnaires to the parents over a 12-month period. Overall direct costs (admission to hospital, diagnostics, surgery, and homecare) were 3 times higher for tracheostomy (€105.523 compared with €33.482, $p=0.02$). Overall indirect costs (absence from work) were almost 5 times higher (€2.543 compared with €543, $p=0.02$). There was a threefold increase in overall total cost/patient (both direct and indirect) for tracheostomy (€108.057 compared with 34.016, $p=0.02$) and 4 times more complications were encountered. This study shows that mandibular distraction osteogenesis in infants diagnosed with Robin sequence costs significantly less and results in fewer complications than tracheostomy, and this contributes to our current knowledge about the ideal approach for infants with Robin sequence and might provide a basis for institutional protocols in the future.


*Keywords:* (Pierre) Robin sequence; Treatment; Tracheostomy; Mandibular distraction osteogenesis; Cost analyses

## Introduction

Mandibular micrognathia leading to glossoptosis and obstruction of the airway are the findings originally described by Pierre Robin in 1923.[1] The sometimes severe obstruction of the upper airway can lead to obstructive apnoea and feeding problems.[2] Non-surgical interventions such as placing the child prone can be applied as a primary treatment and can be useful, particularly in mild cases.[3] However, surgical measures are required in up to 23% of infants with serious respiratory obstruction, which can be challenging.[3] For a long time tracheostomy was considered to be the gold standard. However, the incidence of complications was high and the mean age of decannulation in children with Robin sequence was 28 months, thereby exposing both child and parents to a medical and social burden.[4–7]

The principle of mandibular distraction osteogenesis in infants with Robin sequence is based on lengthening the mandible, so that the base of the tongue is advanced away from the airway, which corrects the supraglottal airway

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obstruction.[3] Numerous reports have been published that illustrate the feasibility of relieving the obstruction, which removes the need for tracheostomy or provides successful decannulation in many cases.[8]

The medical system is in need of treatments that not only provide a good functional outcome, but are also cost-effective. We know of 2 published studies that have compared the costs of tracheostomy and mandibular distraction osteogenesis in infants, yet no distinction was made between infants with isolated or syndromal Robin sequence, and only direct healthcare costs were included.[9,10] The purpose of the present study was to present a comparative cost analysis, including both direct healthcare and indirect (productivity losses) costs for tracheostomy and distraction osteogenesis in infants with isolated Robin sequence treated in a tertiary referral children's hospital in the Netherlands.

## Methods

We retrospectively surveyed the medical records from our hospital from 1 January 1998 to 1 July 2012, and included patients younger than 6 months who had isolated Robin sequence with a supraglottal obstruction that led to respiratory obstruction that could not be treated conservatively, and who were treated by either tracheostomy or mandibular distraction osteogenesis. Until 2007 tracheostomy was routine, but nowadays we prefer mandibular distraction as it causes fewer respiratory complications. Before the intervention all children had a genetic evaluation, monitoring of continuous pulse oximetry for 12 h or more, measurement of blood gases, and flexible fibreoptic airway examination when awake, evaluated by a multidisciplinary team.[11] They were followed up for 12 months, starting on the day the patient started treatment. Ethics committee approval was obtained.

We have listed variables that were of concern. These included direct costs as a result of hospital admission (ward, intensive care unit (ICU) and outpatient clinic, diagnostics (radiology, haematology, microbiology, and virology), surgery (surgical team and devices), and home care (hours of care from a specialised nurse). Other indirect costs related to the parents' absence from work were included. The variables were multiplied with standard reference prices obtained from the Dutch manual for costing research in health care,[12] tariffs from the National Health Authority of the Netherlands[13] or internal cost prices (Table 1). The components of each variable were retrieved by a thorough search of each patient's medical record (Table 2A and Table 2B). The amount of homecare needed and the time the parents spent away from work were obtained retrospectively from a questionnaire (Table 3).

When we calculated duration of absence from work, we assumed that there are 1540 working hours/year.[12,13] The total costs of surgery consisted of the general costs of the team and theatre and the specific costs related to the intervention (equipment). The tracheostomy tubes that we used were Tracoe® 350 neonatology (Tracoe medical GmbH, Frankfurt, Germany) and Shiley™ PED paediatric (Covidien Respiratory and Monitoring Solutions, Boulder, USA). A mean of 4 exchanges/year was used.[14] As this was an outpatient procedure, only the predetermined price for a new tracheostomy tube was calculated. We used a LactoSorb resorbable internal distractor (Walter Lorenz, Inc., Jacksonville, FL) for mandibular distraction osteogenesis.[11]

Table 1
Costs.

| Variable | Reference price (€) per unit |
|---|---|
| *Direct costs* | |
| Hospital | |
| Academic hospital ward | 575.00/day |
| Intensive care unit (ICU) | 2183.00/day |
| Outpatient clinic | 72.00/visit |
| Diagnostic tests | |
| Radiology | 50.00/image |
| Blood | 2.15/test |
| Microbiology | 31.31/test |
| Virology | 26.80/test |
| Surgery | |
| Surgeon | 135.50/h |
| Surgical registrar | 29.45/h |
| Anaesthetist | 135.50/h |
| Anaesthetic registrar | 29.45/h |
| Instrument assistant | 30.50/h |
| Nurse | 30.50/h |
| Mandibular distraction (bilateral) | 1237.26/operation |
| Tracheostomy | 108.26/operation |
| Tracheostomy change[a] | 41.58/change |
| Home care | 35.00/h |
| *Indirect costs* | |
| Absence from work | 30.02/h |

[a] The mean of both tracheostomy tubes was calculated.

Complications during hospital admission and at home were recorded retrospectively. The component that could not be included in the analysis because of lack of data was the number of consultations. Finally the number of visits to the general practitioner, costs of travel to the hospital, ambulance transport, and psychological support were not considered, as they could be prone to recall bias.

Microsoft Excel (Microsoft Inc., Redmond, WA, USA) was used to organise the data, and the significance of differences was assessed with the help of the Mann–Whitney *U* test (IBM SPSS Statistics for Windows 20.0, Amonk, NY, USA, IBM Corp.). Probabilities of less than 0.05 were accepted as significant.

## Results

Sixteen patients required mandibular distraction because of severe respiratory distress caused by a supraglottal obstruction that did not respond to conservative measures. Thirteen met the selection criteria for the present study, but 6 were excluded because they also had additional syndromes and