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REVIEW

A systematic review on the outcome of mandibular distraction osteogenesis in infants suffering Robin sequence

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Abstract
Objectives Mandibular distraction osteogenesis (MDO) has been successfully applied in infants suffering Robin sequence (RS) with severe upper airway obstruction, but no comparative studies for the different types of MDO exist to date. The objective of the current study was to systematically review the published data considering this matter, providing a fundament for protocols and a more conscious treatment strategy for infants with RS in the near future.
Material and methods For the period from January 1966 to January 2012, the Pubmed, EMBASE, and Cochrane Library databases were searched. Abstracts were screened based on predetermined selection criteria. Relevant full-text articles were retrieved. The articles were analyzed on the type of MDO used, preoperative workup, patient characteristics, postoperative outcome, and complications.
Results The search yielded 109 articles. After checking abstracts and full texts on predetermined inclusion and exclusion criteria, 12 studies (four describing external MDO, five internal MDO, and three both types) were extracted for further analyses.
Conclusion Internal MDO seems very feasible in infants suffering RS, minimizing side effects such as hypertrophic scarring, nerve damage, and extensive care needs, although the indications for usage are more limited compared to the external device. Corresponding protocols and long-term outcome studies are needed to make a better comparison and the use and indication of the different types of distraction even more distinct.
Clinical relevance A base for a guideline to support the choice of a designated operative management for neonates with RS is provided, hereby obviating possible complications of the different types of MDO in the future.

Keywords Robin sequence · Mandibular distraction osteogenesis · Micrognathia · Airway obstruction · Review · Outcome

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Introduction

Mandibular micrognathia with associated glossoptosis and airway obstruction is the original triad of symptoms constituting Robin sequence (RS) described by Pierre Robin in 1923 [1]. RS may be an isolated condition, but an associated syndrome such as Stickler's, Treacher Collins, Nager, velocardiofacial, or hemifacial microsomia is present in about 45–80 % of cases [2–6]. An additional cleft palate may exist, but is not a required feature to define the sequence [6–12]. In the literature, the phenomenon is described as Robin complex, Robin anomalad, Pierre Robin syndrome, or Robin sequence [3, 13]. Due to this discrepancy in the definition, the reported incidences vary from 1 per 8,500 [14] to 14,000 [15] births in the general population. Mortality, most commonly due to severe upper airway obstruction leading to obstructive apnea and cardiac problems, ranges from 2.2 to 26 % [16]. To date, several treatment strategies have been proposed, consisting of non-surgical and surgical options. The primary goal is to secure a safe airway in newborns with RS. When there is no life-threatening respiratory obstruction, a conservative approach is applied first. These consist of prone positioning techniques and close monitoring, nasopharyngeal airway, short-term endotracheal intubation, or intraoral devices [6, 16–23]. Although most RS cases can be adequately treated conservatively, up to 23 % of infants have major respiratory obstruction necessitating surgical intervention, which can be challenging for caregivers [24, 25]. Tongue lip adhesion, popularized by Douglas in 1946, has shown its efficacy in protecting the airway in acute respiratory compromise [26–29]. However, potential complications are scarring of the lip, tongue, or salivary glands and dehiscence of the adhesion; secondary procedures may be needed for definite management of the airway [27, 28]. In severe cases, tracheostomy is used as a safe necessary temporary or sometimes even as a long-term measure [16, 30]. The reported mortality rates (most commonly due to accidental decannulation and cannula obstruction) can be up to 6 %, or even higher at a younger age and lower body weight at time of surgery [31]. It is related to a high percentage (43–65 %) of complications such as granulations, tracheocutaneous fistulae, laryngeal/tracheal stenosis, or speech delay [31–33]. Besides, the average age at decannulation in neonates with RS is 28 months, during which an additional substantial social burden on the (grand) parents or caretakers of the child exists to constantly secure the child's airway [34]. Soon after the first widespread clinical use of external mandibular distraction osteogenesis (MDO) by McCarthy et al. in 1992, this technique was also successfully applied in infants suffering RS and was regarded as an alternative corrective treatment for a tracheostomy [35]. Since then, numerous reports have been published demonstrating its feasibility. However, an external device causes external scars and requires a second operative procedure for the removal of the device and screws. The same disadvantage was noticed in the later developed (semi)buried non-resorbable devices, although scars and wound care were minimized [5, 36]. Subsequently, an internal resorbable distractor was presented in 2002 [37]. This technique basically averts the need for a second operation and provides good clinical results [38, 39], which was also demonstrated in our institution [40].

Recently Master et al. [41] gave an extended literature overview of the encountered complications of MDO. However, no clear distinction between the different types of distractors was made, and it was not focused solely on infants with RS. Ow and Chueng [25] presented a well-conducted meta-analyses of the feasibility of MDO, but also did not specify for infants with RS. The purpose of the current study was to comprehensively review the literature regarding both internal and external MDO in infants suffering from RS as this is a vulnerable patient group in which considerable morbidity and even mortality exist at the time a treatment proposal has to be made. At present, no critical systematic review comparing the extensive amount of published case reports about the different types of MDO in this patient group exists. We categorized the outcome per technique, providing a clear summary based on the current knowledge described in the literature and our own experiences regarding MDO. This might help in choosing a designated operative strategy and preventing complications in the future.

Material and methods

Search strategy

A systematic literature search of the PubMed, MEDLINE, and Cochrane databases was performed from January 1966 to January 2012 using specific keywords (Table 1). Duplicates were excluded and abstracts were screened based on predetermined selection criteria. Relevant full-text articles were retrieved and reference lists manually screened for additional articles. Subsequently, the full texts of these articles were critically analyzed (Fig. 1). The search strategy was performed independently by two authors (E.P. and F.S).

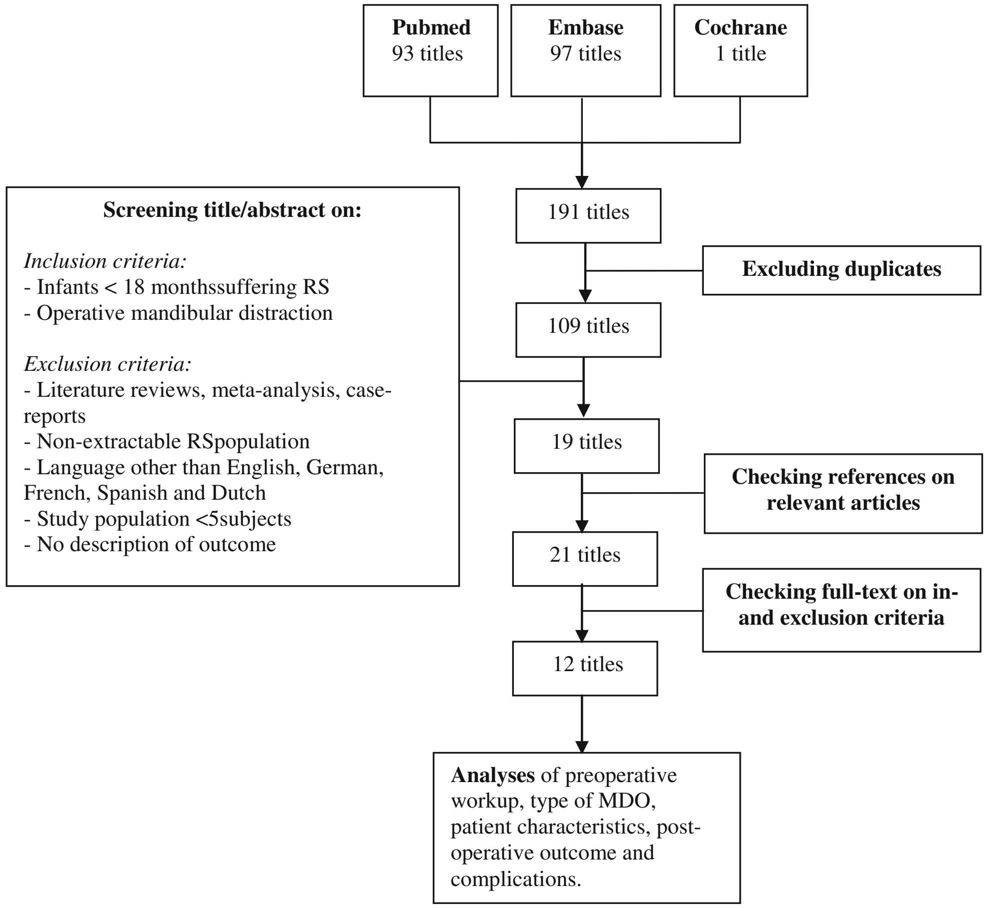
Inclusion and exclusion criteria

Prospective and retrospective case series, describing the outcome after external or internal mandibular distraction using resorbable or non-resorbable distraction device, in more than five infants aged <18 months suffering from RS

Table 1 Keywords used for the search of the three databases

PubMed	("mandibular"[Title/Abstract] OR "mandible"[Title/Abstract] OR "jaw"[Title/Abstract] OR "jawbone"[Title/Abstract] OR "chin"[Title/Abstract]) AND ("distraction"[Title/Abstract] OR "extend"[Title/Abstract] OR "extension"[Title/Abstract] OR "enlargement"[Title/Abstract] OR "distention"[Title/Abstract] OR "expansion"[Title/Abstract] OR "osteogenesis"[Title/Abstract]) AND ("pierre"[Title/Abstract] OR "robin"[Title/Abstract] OR "sequence"[Title/Abstract])
EMBASE	(mandibular:ab,ti OR mandible:ab,ti OR jaw:ab,ti OR jawbone:ab,ti OR chin:ab,ti) AND (distraction:ab,ti OR extend:ab,ti OR extension:ab,ti OR enlargement:ab,ti OR distention:ab,ti OR expansion:ab,ti OR osteogenesis:ab,ti) AND (pierre:ab,ti OR robin:ab,ti OR sequence:ab,ti)
Cochrane	(mandibular:ti,ab OR mandible:ti,ab OR jaw:ti,ab OR jawbone:ti,ab OR chin:ti,ab) AND (distraction:ti,ab OR extend:ti,ab OR extension:ti,ab OR enlargement:ti,ab OR distention:ti,ab OR expansion:ti,ab OR osteogenesis:ti,ab) AND (pierre:ti,ab OR robin:ti,ab OR sequence:ti,ab)

Fig. 1 Flowchart search strategy



were included for further assessment. Studies consisting of a mixed group of patients, in which those diagnosed with RS could not be extracted from the whole study group to analyze the outcome separately, were excluded. If there was double reporting of patients from the same center in different publications, the article describing the largest group was included. Articles that did not meet the selection criteria (description of preoperative workup, type of MDO used, patient characteristics, postoperative outcome, and complications) were likewise excluded.

Results

The literature search retrieved 191 titles. Following the criteria as described in detail in Fig. 1, the search strategy finally yielded a total of 12 articles that were included for critical analyses. Four papers described the use of an external distractor [28, 42–44], three of an internal non-resorbable distractor [36, 45, 46], another three studies [5, 47, 48] described the use of both kinds of distracters, and two of an internal resorbable distractor [38, 40]. There was no disagreement between the two assessors regarding the inclusion process.

A total of 212 patients underwent MDO over the period 2004–2012. Of these, 82 % suffered isolated RS, 8 % Stickler's syndrome, 2 % Treacher Collins, 1 % Opitz Syndrome, and the rest suffered from varying syndromes. A full genetic evaluation was performed in all patients routinely. A cleft palate was seen in 79 % of cases. The mean age of MDO cases varied from 8.6 weeks in external MDO, 9.6 weeks in internal non-resorbable MDO, and 8.3 weeks in resorbable MDO. Preoperatively, a nasal fiber-optic airway investigation was performed in all studies to rule out any other possible causes of airway obstruction. In some studies, additionally, bronchoscopy was done [28, 45, 48]. Except for one study [36], polysomnography was performed to rule out the presence of central apneas or as a parameter to compare pre- and postoperative outcomes. Two others did not perform this if obvious upper airway obstruction was present even while the infant is awake [40, 44]. Feeding status and observations, oxygen saturation monitoring, arterial blood gas measurement, and radiographic imaging (cephalometry and/or 3D CT scans) were always carried out; a multidisciplinary team approach was also described in all articles (Table 2). The mean durations of the distraction process were 17 days for the external devices, 11 days for the internal non-resorbable devices, and 8.5 days for the resorbable devices. The mean total amounts of distraction were 11.6 mm (external), 17.3 mm (internal non-resorbable), and 18.3 mm (resorbable). The mean consolidation period amounted respectively 5.5, 7, and

Table 2 Patient characteristics

Article	Type of MDO	Patients receiving MDO (n)	Diagnosis	Cleft palate (n)	Mean age at MDO (weeks)	Preoperative investigations	Indication for distraction
Morovic et al. [43]	External	23/31	Isolated RS (n=27) Stickler's (n=4)	90 % (21)	10 (0.7–36)	Genetic evaluation, preoperative team evaluation, feeding observations and nutritional status, lateral cephalogram, naso-fiber-optic airway examination, O_2 saturation monitoring, polysomnography (on indication)	Micrognatia with severe respiratory difficulty, interruption of the air column on lateral cephalogram, no signs of laryngeal associated pathology on naso-fiber-optic examination+ frequent desaturations (<80 %) not corrected by conservative measures needing intubation; (2) micrognathia+presence of a tracheotomy; (3) Micrognathia+insufficient growth and/or malnutrition and/or great irritability and/or aberrant polysomnography; (4) Micrognathia with acute respiratory difficulty after palatoplasty
Denny et al. [28]	External	11	Isolated RS (n=3) Unidentified (n=1) Stickler's (n=5) Freeman Sheldon (n=1) VCF (n=1)	91 % (10)	2.6 (0.5–6.4)	Genetic evaluation, preoperative team evaluation, nasopharyngoscopy, CT and 3D CT scan, flexible fiber-optic bronchoscopy, O_2 saturation monitoring, length and weight data, polysomnography (only the last 7 patients)	Upper airway obstruction due to tongue posture caused by a small mandible as a primary source of obstruction, as evidenced on radiologic evaluation and nasopharyngoscopy, no tracheo/laryngomalacia or other causes compounding tongue ptosis (e.g., circular pharyngeal collapse) and severe refractory obstruction (desaturations<70 %) otherwise requiring neonatal tracheostomy for airway control. Traditional measures (tongue–lip adhesion/prone positioning/nasopharyngeal airway intubation) failed.
Scott et al. [44]	External	19	Isolated RS (n=14) Stickler's (n=2) Opitz (n=1) Arthrogryposis (n=1) Catel Manzke (n=1)	?	4.8 (0.7–12)	Genetic evaluation, preoperative team evaluation, O_2 saturation monitoring, airway endoscopies, radiographic imaging, polysomnography (on indication)	Children diagnosed with RS in who should failure to thrive despite conservative measures and tracheotomy and/or G-tube placement was the only other viable alternative.
Monasterio et al. [42]	External	18	Isolated RS (n=17) Orofacio-digital (n=1)	72 % (13)	17 (1.1–21.4)	Genetic evaluation, preoperative team evaluation, O_2 saturation monitoring for 8 h, blood gasses, nasopharyngoscopy, radiographic imaging, esophageal pH studying, polysomnography barium videofluoroscopy	Micrognathia, glossoptosis and upper respiratory obstruction, snoring and respiratory obstruction which were observed in supine position were corrected in the ventral position and cyanosis during feeding requiring >20 min for ingestion
Shen et al. [36]	Internal non-resorbable	6	Isolated PRS (n=6)	100 % (6)	1.9 (1–3)	Genetic evaluation, preoperative team evaluation, O_2 saturation monitoring, perinatal feeding history, 3D CT scan and lateral cephalogram, airway endoscopy	Mandibular retrognathia, glossoptosis and cleft palate formation+(1) traditional management (prone positioning, nasopharyngeal airway intubation) failed and the perceived need for long-term respiratory support, result of preoperative O_2 saturation of approx. 40 % in prone position+(2) distance post-pharyngeal wall to lingual root on lateral cephalogram<3 mm
Mohamed et al. [46]	Internal non-resorbable	11	?	100 % (11)	18 (8–22)	Genetic evaluation, preoperative team evaluation, O_2 saturation monitoring, feeding observations, lateral cephalogram, O_2 saturation monitoring, polysomnography, CT scan and 3D CT scan	Retromicrognathia, glossoptosis and cleft palate and symptoms of OSAS, unable to control airway during feeding, high respiratory disturbance rates and O_2 saturation ranged 75–85 % on polysomnographic, not responding to conservative measures (nasopharyngeal airway or gloosopexia)

Table 2 (continued)

Article	Type of MDO	Patients receiving MDO (n)	Diagnosis	Cleft palate (n)	Mean age at MDO (weeks)	Preoperative investigations	Indication for distraction
Hong et al. [45]	Internal non-resorbable	5	Isolated RS (n=2) 4p deletion (n=1) Stickler's (n=1) Otopalatodigit (n=1)	100 % (5)	9.1 (4–13.4)	Genetic evaluation, preoperative team evaluation, flexible pharyngolaryngoscopie, bronchoscopy, imaging of the craniofacial skeleton, polysomnography videoflueroscopic swallow tests (modified barium swallow) and upper GI series (barium swallow)	Infants who were intubated at birth and later failed extubation and/or presented with significant O_2 desaturation with signs of respiratory distress despite conservative measures, such as positioning
Mandell et al. [48]	Internal (n=?); external (n=?)	8	Isolated PRS (n=7) Beal (n=1)	38 % (3)	2.5 (1.1–32)	Genetic evaluation, preoperative team evaluation, feeding status and observation, 3D CT scan, flexible nasolaryngoscopy and direct laryngoscopy, polysomnography, bronchoscopy, O_2 saturation monitoring	Micrognathia and respiratory obstruction due to tongue base collapse, severe enough to require admission to the NICU and whose airway obstruction could not be controlled with conservative measures (positioning and nasal airway), no presence of a second airway lesion, central sleep apnea and/or severe untreated GERD, and tracheotomy was recommended based on a consensus decision by multidisciplinary team.
Genecov et al. [5]	Internal (n=33); external (n=34)	67	Isolated RS (n=64) Treacher Collins (n=3)	81 % (67)	56 (0.7–288)	Genetic evaluation, preoperative team evaluation, O_2 saturation monitoring, flexible upper and lower airway endoscopies, lateral X-ray of the neck, pH probe, CT scan head and cervical spine, polysomnography	Airway obstruction associated with micrognathia, glossoptosis, gastroesophageal reflux, micro-aspiration, swallowing abnormalities and failure to thrive with O_2 saturations<92 % on supplemental O_2 without improvement by prone positioning and no response on NP tube and/or nasal continues positive airway pressure and no other signs of airway obstruction seen on panendoscopy, neurologic impairment (lack of airway protection) and untreated GERD with (epi)glottic edema
Wittenborn et al. [47]	Internal (n=13); external (n=4)	17	Isolated RS (n=14) Stickler's (n=2) Treacher Collins (n=1)	100 % (17)	4 (0.7–17.1)	Genetic evaluation, preoperative team evaluation, feeding observations and nutritional status, CT scan and 3D CT scan, naso-fiber-optic airway examination, polysomnography, polygraphic studies: heart rate, respiratory rate, chest wall impedance, nasal airflow, EMG, O_2 saturation monitoring	Infants with intractable upper airway instability caused by glossoptosis secondary to mandibular micrognathia and cleft palate, in the absence of airway pathology beyond the glossoptosis, not responding to conservative measures (prone positioning, chest rolls and/or nasopharyngeal tubes) and (1) in direct need of a tracheotomy or (2) showing increasing respiratory problems and failure to thrive after observation
Burstein et al. [38]	Internal resorbable	15	Isolated RS (n=12) Stickler's (n=1) Opitz (n=1) Bilateral craniofacial microsomia (n=1)	100 % (15)	12 (1–44)	Genetic evaluation, preoperative team evaluation, O_2 saturation monitoring, lateral and anterior posterior cephalogram, awake flexible fiber-optic airway examination or direct laryngoscopy and bronchoscopy in already intubated patients, polysomnography (when clinically stable enough)	Severe life-threatening airway obstruction secondary to retromicrognathia with clinical symptoms

Table 2 (continued)

Article	Type of MDO	Patients receiving MDO (n)	Diagnosis	Cleft palate (n)	Mean age at MDO (weeks)	Preoperative investigations	Indication for distraction
Breugem et al. [40]	Internal resorbable	12	Isolated RS (n=8) Stickler's (n=2) Suspected Stickler's (n=2)	100 % (12)	4.6 (1.6–13.4)	Genetic evaluation, preoperative team evaluation, continuous O_2 saturation monitoring, blood gas evaluation, lateral and anterior posterior cephalogram, 3D CT scan, awake flexible fiber-optic airway examination, polysomnography (on indication)	Infants with glossoptosis, micrognathia and airway compromise which could not be treated with conservative measures (prone positioning, nasal continuous positive pressure) and were considered candidates for tracheotomy, with no other causes of airway obstruction seen on endoscopy (tracheomalacia, stenosis) besides the glossoptosis and saturation was <90 % for >5 % of 12 h observation

4 weeks. The mean distraction rate varied from 1 mm/day in the external devices up to 2 mm/day for the resorbable devices. Extubation or decannulation data were missing in some articles and ranged respectively 3–15, 4-6, and 5–7.5 days postoperatively. Drive screws from the resorbable devices were removed ambulatory without sedation and the external devices under sedation, but for the internal devices, mainly general anesthesia was necessary [45, 46] (Table 3). All reviewed articles described an overall positive outcome, avoiding a tracheostomy or obtaining a successful decannulation in 82 % [47], 89 % [44], and 94 % [5] up to 100 % [28, 36, 38, 40, 43, 45, 46, 48, 49] of cases. Normal oral feeding after MDO was possible in 86 % [48] and 91 % [5] up to 100 % [28, 36, 40, 45, 46] of cases, and if the growth of the infant was registered, there was normalization in the growth curve seen in almost 100 % cases after 1 year [28, 43] (Table 4). Cumulative complications described are listed in Table 5.

Discussion

Distraction osteogenesis was introduced by lengthening of the femur by Codvilla in 1905 [50] and the tibia by Abbott in 1927 [51], although it was not until the 1940s that the use in the lower extremities was truly popularized by Illizarov [52] and De Bastiani et al. [53]. Application of the technique in the craniofacial skeleton should be credited to German craniofacial surgeons Wassmund [54] (for clinical advancement of the maxilla in a patient with hypoplasia) as early as 1926 in Berlin and to Rosenthal and Sonntag [55] (for bone lengthening of the mandible in a micrognathic patient) in 1927. MDO, a term introduced by Rosenthal, seemed to be forgotten worldwide until 1972, when an experimental report in a canine mandible was presented by Sneyder et al. [56]. This was followed by a clinical report of McCarthy et al. in 1992 [35], describing a rigid external device for distraction of the mandible in congenital deformities. Since then, numerous reports have been published, demonstrating the feasibility in relieving airway obstruction, preventing tracheotomies, or providing a successful decannulation in many cases [25, 57–60]. Currently, there are two main types of devices: external and internal distractors. Internal distractors can be subdivided into non-resorbable and resorbable distractors. One of the advantages of internal distractors is the lack of a cumbersome external device during distraction and the consolidation period. This tends to make the process more acceptable for the parents, offering the possibility for the mother to breastfeed and maintain expander integrity. Also, there is a smaller risk of pin-associated infections compared to external MDO where pin site hygiene can be challenging. External scars are less notable due to their location under the mandibular ramus, hypertrophic scarring is less common, and there is less risk to damage the marginal mandibular branch of the facial nerve [61–63]. The primary advantage of external distractors is the ability to use multivector movement performing multiplanar distraction, which can be adjusted during the distraction phase to accommodate to mandibular asymmetries or irregularities [63]. The unidirectional movement of internal distractors requires a more meticulous planning of osteotomies and vectors and does not allow fine adjustments of mandibular segments to correct any occlusal disharmony that occurs during the distraction process [64]. Curvilinear devices have been developed, trying to obviate this problem [65]. Still, both internal and external MDO will require a second operative procedure for removal of the hardware, which is prevented by the use of an internal resorbable distractor. Resorbable distraction has been widely used in craniofacial surgery for several years [66]. The application in infants with RS was first presented in 2002 by Burstein et al. [37]. They illustrate promising outcomes and address the benefits of this one-stage surgical procedure and the clinical applicability especially suited for infants. Ow and Cheung [25] present an extended meta-analyses of MDO. However, they do not differentiate between the different types of distractors or describe results for infants in particular. In the present study, it was the aim to review the distraction features, results, and possible complications per type of distraction and summarize the possible differences.

In all the reviewed articles, independent of the type of distractor used, MDO was only performed when conservative measures failed. This was determined by a multidisciplinary team after a range of preoperative investigations (Table 2). It is an important fact to emphasize as the usage of nasopharyngeal intubation [67] or intraoral devices such as a palatal plate with pre-epiglottal extension [20, 23] reveals pleasing clinical results, especially in mild forms of respiratory distress. Considering distraction features, it was noticed that the duration of the distraction process of the internal resorbable device was twice as short compared to the external device (Table 3). This could be explained by the faster distraction rate, which also led to a quicker discharge from the hospital compared to external MDO [38, 40]. Besides, in external MDO, the distraction process sometimes started only on the third postoperative day [43, 49] compared to mainly the first day in internal MDO. Notably, in internal MDO, the mean total amount of distraction was also longer (17.3 and 18.3 in internal vs. 11.6 in the external device). Extubation or decannulation time was not much different between the different types of MDO, although not all articles specified this. The consolidation period was also generally corresponding for the different types of distractors. Some authors provoked a slight overcorrection of the mandible compared to the maxilla to indicate the end of the distraction process [36, 47, 49] as others objectified a normal horizontal position of the tongue [28, 48] or an anatomic maxilla–mandibular proportion [40, 45]. Overcorrection is performed to compensate for the regenerative contraction that can occur due to a possible decrease in growth capabilities [36]. Morovic [43] saw a disproportional overprojection of the mandible during the first year of age, corrected at 1 year in 70 % and at 18 months in the remaining 30 %.

The complications encountered consisted mainly of local infection of the skin [36, 38, 40, 43, 45–47, 49], which did not lead to any delay in the distraction process. Infections of the skin were remarkably more often seen in resorbable distractors, which might possibly be due to a reaction caused by the degradation process. All infections healed with topic antibiotic ointment. With regard to the non-internal resorbable devices, Genecov et al. [5] saw an almost twice as high infection rate in external compared to internal MDO (8.8 vs. 4.5 %). Secondly, facial nerve problems were encountered in both the external and internal approaches and were mainly transient when due to traction on the nerve during the placement of the distractor [5, 44, 46]. Long-term injury of the marginal mandibular branch of the facial nerve leading to asymmetric movement of the lower lip at the corner of the mouth was associated more often with the external approach as the nerve is not visible during placement of the distractor [5, 44]. No nerve paralysis was seen in the internal resorbable distraction groups [38, 40]. Device failure leading to replacement was the third most common complication, seen both in external and internal non-resorbable MDO, but not in resorbable MDO. Hypertrophic scarring was only seen in the external devices. In two articles, no complications were encountered [28, 36] (Tables 4 and 5). An overall smaller amount of complications was seen in internal MDO compared to external MDO (Table 5). Some authors described unsuccessful decannulation or long-term need of a tracheostomy during MDO. Since long-term problems are mainly encountered in the syndromal, more complex cases, MDO should be strongly reconsidered in this patient group as a variety of underlying potential factors (neurologic dysfunction, persistent supraglottic obstruction, or temporomandibular joint ankylosis) might not allow decannulation [44, 48]. Up to now, long-term results of only external MDO are described [28, 44]. Especially the size and shape of the distracted mandible and outgrow of the teeth is of great concern [44]. Denny and Amm [28] have demonstrated after 5 years of follow-up that the mandible was growing appropriately in all patients receiving external MDO. Scott et al. [44], with a medial follow-up of 67 months, show that in 5 % of the patients there exists a residual open bite deformity and in 21 % long-term tooth loss or malformation after external MDO. Teeth damaged were the first, second, and premolars, likely related to the location of the mandibular osteotomy. They prefer an osteotomy posterior to the tooth buds to prevent tooth loss, however not through the mandibular angle, which can be challenging. The same applies for the

Table 3 Distraction features

Article	Type of MDO	Start (postop day)	Mean duration of distraction (days)	Indicator of end of distraction	Distraction rate (mm)	Mean total amount of distraction (mm)	Consolidation period (weeks)	Removal of MDO	Extubation or decannulation (days)
Morovic et al. [43]	External	3	21	After resolving the obstruction measured by nightly saturation and/or polysomnography	1/day	18–25	Age<6 months: 4; Age>6 months: 6 controlled by lateral cephalometry	Ambulatory	15
Denny et al. [28]	External	1	14	Observed by correction of the tongue from initial vertical to a physiologically normal horizontal posture	Days 1–3: 2/day; From day 3: 1/day	12.4 (8–15) extracted from Denny [60]	4	Sedation	3–5
Scott et al. [44]	External	?	?	?	?	?	4–8	?	?
Monasterio et al. [42]	External	3	15	Until overcorrection was evident	1/day	12 (7–19)	6–8	?	?
Shen et al. [36]	Internal non-resorbable	1	15	When max. length of distractor was reached (20 mm) or mandibular gum line was 0–2 mm in front of maxillary gum line	Days 1–5: 1.2/day; From day 6: 1/day	16 (9–21)	4	Sedation	4–6
Mohamed et al. [46]	Internal non-resorbable	1	?	?	0.5/twice a day	18 (15–23)	10	General anesthesia	?
Hong et al. [45]	Internal non-resorbable	1	9–12	Until no maxilla-mandibular discrepancy is left	1.5–2.0/day	18 (15–20)	6-8	General anesthesia	?
Mandell et al. [48]	Internal and external	1–2	12 (8–18)	When airway obstruction was corrected, confirmed with flexible laryngoscopy or conversion of tongue orientation from vertical to horizontal position	0.6/three times a day	23 (15–23)	6	Sedation	5.6 (1–8)
Genecov et al. [5]	Internal and external	1	19.4	Once central end-to-end occlusion was achieved	1/day	22 (10–32)	10.4	External: often office procedure; Internal: ?	?
Wittenborn et al. [47]	Internal and external	1	7.6 (6–15)	When mandibular alveolar arch was 1 mm ventral to the maxillary alveolar arch; maximum activation of the distractor was reached or non-progression of the distractor	1–1.2/twice a day	12–15	4–6	External: office procedure; Internal: outpatient procedure	7.6 (6–15); after completion of the distraction
Burstein et al. [38]	Internal resorbable	2	9	After maximal technical distraction was achieved (based on length of the drive screws)	2/day	18 (15–20)	4	Ambulatory without sedation	5–7
Breugem et al. [40]	Internal resorbable	1.5–2	8 (6–9)	Until mandibular alveolus was in normal position compared to the maxilla or maximal technical distraction was achieved	1/twice a day	18.5 mm (16–22)	4	Ambulatory without sedation	7.5 (5–11)

Table 4 Outcome

Article	Type of MDO	General outcome	Mean follow-up (months)	Complications described
Morovic et al. [43]	External	Complete relief in respiratory symptomatology in 100 %; early decannulation in 100 %; normalization of growth curve in 100 % ; proportional mandibular growth at 18 months in 100 %	18	13 % transient local infection of the skin (n=3) 9 % extrusion of the nails (n=2) 9 % hypertrophic scar (n=2)
Denny et al. [28]	External	Normal postoperative polysomnography after 1 week–1 month in 100 %; normal oral feeding in 91 % after 45 days and in 100 % after 1 year; growth above the 50th percentile in 91 %	60	No complications
Scott et al. [44]	External	Avoidance of a tracheotomy in 89 % (n=17); 90 % (n=17) outcome score of ≥8/10 (“intermediate–good”)	67 (36–145)	21 % long-term tooth loss or malformation (n=4) 16 % hypertrophic scars (n=3) 16 % long-term injury of the marginal mandibular branch of the facial nerve (n=3) 16 % G-tube-dependent (n=3) 5 % additional MDO due to failure of mandibular growth and relapsing upper airway obstruction (n=1) 5 % inability to decannulate (n=1) 5 % residual anterior open-bite deformity (n=1)
Monasterio et al. [42]	External	Disappearance of gastroesophageal reflux and sleep apnea in 100 %; mean oxygen saturation 93 % (range, 89–96 %); disappearance of abnormal tongue movements and barium stasis in the pharyngeal recess and trachea in 100 %; normal pharyngeal transit time in 100 %	2–4	28 % transient local infection of the skin (n=5) 6 % incomplete osteotomy requiring second operative procedure (n=1)
Shen et al. [36]	Internal non-resorbable	No supplemental oxygen after 20 days postop in 100 % (n=6), no additional treatment or surgery needed in 100 % (n=6); full oral feeding after 1 month in 100 % (n=6)	6	No complications
Mohamed et al. [46]	Internal non-resorbable	Uneventful decannulation, removal of tongue stitches or nasopharyngeal airway (100 %); mean respiratory index 1 (range, 0–2); mean oxygen saturation 97 % (range, 95–99 %); normal oral feeding, no breathing problems, and normal growth after 6–12 months (100 %)	12–24	27 % transient mild infections of the skin (n=3) 9 % unilateral incomplete osteotomy (n=1) 9 % transient unilateral mild weakness of marginal branch of facial nerve (n=1)
Hong et al. [45]	Internal non-resorbable	Avoidance of tracheotomy and other airway interventions (including supplemental oxygen) in 100 %; no monitoring or other home care measures on discharge in 100 %; improvement in swallowing function and reflux disease and full oral feeding in 100 %	18	40 % local erythema and tenderness (n=2)

Table 4 (continued)

Article	Type of MDO	General outcome	Mean follow-up (months)	Complications described
Mandell et al. [48]	Internal and external	Full relief of upper airway obstruction and discharged home at 7 days (range, 2–62 days) after MDO (88 %); no regular home monitoring in 100 %; normal oral feeding in 86 %	13 (7–16)	12.5 % G-tube-dependent (n=1)
Genecov et al. [5]	Internal and external	Decannulation for 1 year after MDO or prevention of an eminent tracheotomy in 94 % (n=63); successful swallowing in 91 % (n=61); reduced postop respiratory disturbance index in 97 % (n=65)	?	Local infections of the skin: 4.5 % in internal MDO (n=3) and 8.8 % in external MDO (n=6) Replacement related to device failure: 3 % in internal MDO (n=2) and 10.2 % in external MDO (n=7) Temporary asymmetric movement of the depressor anguli oris muscle: 4.5 % in internal MDO (n=3) and 4.5 % in external MDO (n=3) 3 % failed decannulation (n=2)
Wittenborn et al. [47]	Internal and external	Successful long-term outcome in airway stabilization after extubation in 82 % (n=14)	16.5 (8–48)	Need for tracheotomy: 25 % in external MDO (n=1) and 8 % in internal MDO (n=1)
Burstein et al. [38]	Internal resorbable	Avoidance of a tracheotomy in 100 % (n=15); early decannulation in 93 % (n=14), 7 % requirement of fundal plication due to severe gastroesophageal reflux (n=1)	24	27 % transient local infections of the skin (n=4)
Breugem et al. [40]	Internal resorbable	Discharge (mean, 17 days postoperatively.; range, 11–27) without any nasal continuous pressure in 100 % (n=12); normal oral feeding in 54 % (n=6) at discharge and in 36 % (n=4) within 4 weeks postoperatively	32 (13–56)	8 % transient local infections of the skin (n=1) 8 % extrusion of the distraction screw (n=1)

pins, which preferably are not placed in the tooth buds. Careful planning and modeling may limit complications, although optimal vectors for distraction should be achieved as well, making it sometimes a difficult consideration [68]. Three-dimensional CT scan to locate the foramina of the inferior alveolar nerve and the distal tooth buds in the mandibular bodies [36] or an acrylic model of the skull [39] might be helpful preoperatively. However, in the presence of a life-threatening situation, early intervention is often essential, and disadvantages like tooth injury should be weighed against the benefits of preventing an acute tracheostomy [68, 69]. Long-term results of internal distraction, both resorbable or non-resorbable, are not yet available.

The preference for a type of distraction remains questionable as both have their advantages and indications, as outlined above. Three articles described the use of both kinds of distractors [5, 47, 48]. Wittenborn et al. [47] prefers the internal distractor due to less chance of device failure and simplified care during the distraction and consolidation process. Genecov et al. [5] depended their choice on the availability of the distractors and the experience of the team. Indications for the use of external distractors included the need for large advancements (>20 mm) and multidirectional vectors and the presence of adequate bone stock volume. Internal distraction was preferably used in infants younger than 1 year and a planned single-vector distraction. In their series, the internal devices showed minimal scarring, the incidence of pin site infection was lower, and breastfeeding was easier. Mandell and co-workers [48] also preferred the internal method when feasible because of less change of device dislodgment, no visible hardware allowing earlier return to school or daycare, reduced parental anxiety, and less visible facial scarring. The available small internal resorbable distractors have been proven to be very suitable for airway management in infants [38–40]. With the development of curvilinear internal devices [65, 70, 71] and even the preliminary use of bone morphogenetic proteins to accelerate bone healing during the distraction process [39, 72], this is a

Table 5 Complications

	External device (n=109)	Internal device (n=95)	
		Non-resorbable (n=68)	Resorbable (n=27)
Local infection of the skin	12.8 % (14)	11.8 % (8)	18.5 % (5)
Nerve damage	5.5 % (6)	5.9 % (4)	–
Device failure needing replacement	6.4 % (7)	2.9 % (2)	–
Hypertrophic scars	4.6 % (5)	–	–
Tooth loss or malformation	3.7 % (4)	–	–
G-tube-independent[a]	2.8 % (3)	–	–
Incomplete osteotomy	1.8 % (1)	1.5 % (1)	–
Failed decannulation/need for tracheotomy[a]	1.8 % (2)	1.5 % (1)	–
Extrusion of the nail	1.8 % (2)	–	3.7 % (1)
Need for second operative measure	1.8 % (2)	–	–
Other	0.9 % (1; open-bite deformity)	–	–
Total complications	43 % (47)	24 % (16)	22 % (6)
No complications	57 % (62)	76 % (52)	78 % (21)

[a] 12.5 % G-tube-dependent (n=1) in Mandell et al. [48] and 3 % failed decannulation (n=2) in Genecov et al. [5] are complications which are not included as no difference could be made between external and internal MDO

promising and upcoming technique. Current results are pleasing, although long-term data considering outgrow of the mandible are lacking [38–40].

Even when an additional temporary tracheostomy or endotracheal intubation is mandatory besides MDO to relieve acute severe upper airway problem, all the reviewed articles illustrated that most patients could be decannulated or extubated successfully before discharge. It is imperative to remember that in our quest to find the appropriate surgical treatment, glossopexy and tracheostomy may be lifesaving in the acute phase, but do not correct the micrognathia, which is the basic anatomical pathology of RS [28, 49]. After MDO, a quicker rehabilitation associated with less risks and long-term comorbidities is possible [27, 28, 31–33]. Moreover, homecare is less cumbersome and the total care is less expensive compared to a tracheostomy [73, 74]. Some authors support the "growth catch-up theory" as an argument not to perform MDO [75, 76]. However, when conservative measures fail, a more aggressive approach must be selected at that sudden moment. In contrast to tracheotomies and tongue–lip adhesions which are considered as a transient intervention pending an eventual normal intrinsic normal outgrow of the mandible, MDO is a more definite treatment option and a safe fundament for further growth of the mandible [28, 38]. Besides, although some patients may outgrow their micrognathia without intervention, rarely does the mandible reach normal values for size matched with normal infants, making a definite treatment option for infants suffering RS more essential [77]. Nevertheless, patients that have received MDO may still need a second operative correction of the mandible later in life as long-term studies (i.e., longer than 5 years) are not yet available and some form of relapse has been described in up to 64.8 % of cases after MDO in general [41]. With long-term results for MDO in infants with specifically RS being unavailable, certain questions still arise, such as the duration of the distraction and the timing of the removal of the device. In the resorbable device, the relation between the speed of degradation of the plate and the presence of a possible (long-term) relapse or outgrow problem still has to be elucidated. Besides, although all articles included in the current study handled mainly the same fundamentals in starting MDO only when conservative measures failed, no strict corresponding protocols suggesting a clear indication were used. Therefore, there is potential for bias, both in this study and in other reports [48]. Also, the moment when distraction is finished remained somewhat indistinct in some of the reviewed articles. Some used clinical parameters (e.g. optimization of saturation) whereas others used more technical (e.g. maximal length of distractor was reached) or anatomic aspects (e.g. overcorrection of mandible compared to the maxilla or normal position of the tongue) as indicators of the end of distraction. This is an important fact as the chance of a relapse might depend on the amount of distraction, but which cannot be well compared now. Finally, as not all articles specified the results for the syndromal and non-syndromal RS patients, it is difficult to find an overall difference in outcome between these groups. It is known that there is a higher mortality rate in children with associated anomalies: 22.8 % compared with 5.9 % for those with isolated RS [6]. This should be regarded in choosing a treatment strategy for this patient group as a different approach might be necessary. More specific guidance for MDO in infants with RS consisting of standardized indications, patient selection, pre- and postoperative evaluation, and distraction processes need to be established [9, 78]. Besides, prospective trials comparing different treatment strategies are necessary and might reveal valuable knowledge for the development of an "ideal approach" [23, 67].

With regard to the reviewed literature and our own experience, we would advise a non-surgical approach as a primary measure in all cases. When there is a life-threatening respiratory obstruction—and any other causes and comorbidities are thoroughly investigated by the aforementioned investigations in a multidisciplinary approach—we suggest performing MDO. In young infants (i.e., younger than 6 months), an internal (resorbable) distractor has shown promising results with regard to immediate airway obstruction relief and also parental acceptance and tolerability in home care. In the older or multi-complex cases, an external distractor might provide benefits over an internal device. Using guided surgery by means of preoperatively planned surgiguides [79] or navigation [80] could eliminate the need for external devices in complex cases and support the utilization of curvilinear internal devices. Additional treatment (for example, nasogastric tube feeding) should be started during the distraction process to prevent any further growth retardation. To obviate the lack of comparable data, especially for the amount of distraction needed to resolve the respiratory obstruction, we would also recommend measuring the amount of sagittal discrepancy between the maxillary (point A) and mandibular arch (Pogonion) before and after distraction in all future studies. This will contribute to developing designated treatment algorithms and might provide a fundament for protocols in the near future.

Conclusion

When conservative measures fail, MDO proves to be an appropriate and safe intervention in infants and obviates the side effects seen with tracheostomy and tongue–lip adhesion. This review suggests that the internal device seems very feasible in infants with RS, minimizing side effects such as hypertrophic scarring, nerve damage, and extensive care needs, although the indications for usage are more limited compared to the external device. Corresponding protocols and long-term outcome studies are needed to make a comparison between the different types of distraction possible and their use and indication more distinct.

Conflict of interest The authors declare that they have no conflict of interest.

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