


The Reproducibility of the Jaw Index in the Measurement of Healthy Newborns

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Abstract

Objective: Establish the reliability of the jaw index to objectify the relationship between the maxilla and mandible in healthy newborns.

Design: Cohort study.

Setting: Tertiary setting.

Patients: A total of 52 healthy newborns were included to detect an inter and intraclass correlation coefficient (ICC) of 0.8 with a 95% confidence interval (95% CI) of width 0.3. Inclusion criteria were children born full term without respiratory or feeding problems, and without congenital malformations or facial deformities due to birth trauma. Uncooperative patients were excluded.

Interventions: The jaw index, a measuring tool for objectifying micrognathia in children suspected of having Robin sequence, was used. An ICC of greater than 0.8 was considered clinically relevant.

Main Outcome Measure(s): Primary outcomes are the reliability of the jaw index expressed as interclass correlation coefficient and ICC. Secondary outcomes are the mean jaw index and mean length of the mandible, maxilla, and the alveolar overjet.

Results: An interclass correlation coefficient of 0.74 (95% CI: 0.49-0.86) and an ICC of 0.81 (95% CI: 0.66-0.89) were found. The mandible had an average length of 162.6 mm (standard deviation [SD] 11.1), the maxilla 168.7 mm (SD 9.4), the alveolar overjet 2.0 mm (SD 0.60), and the mean jaw index was 2.1 (SD 0.64).

Conclusion: The jaw index is a consistent instrument between different observers as well as for one observer measuring consecutively in the same child, to objectify the size of the lower jaw compared to that of the upper jaw in healthy newborns.

Keywords

Robin sequence, jaw index, reliability

Introduction

Robin sequence (RS) is a congenital malformation first described by Pierre Robin in 1923 and is defined by a triad of micrognathia, glossoptosis, and upper airway obstruction (UAO) (Robin, 1923; Robin, 1994).

In later publications, cleft palate and gastroesophageal reflux were added as aggravating factors to the definition of RS (Robin, 1923; Dudkiewicz et al., 2000; Baujat et al., 2001; Wagener et al., 2003). Since then, the definition of RS has been under constant evaluation and debate as numerous other modified definitions have been proposed (Basart et al., 2015). Due to the heterogenic definitions used, comparison between case series was often unreliable. So, during the Robin Sequence Consensus Meeting in 2014 in Utrecht, the Netherlands, an

international Clinical Consensus Report was developed, which established micrognathia, glossoptosis, and UAO as the sole triad of symptoms that characterize RS (Breugem et al., 2016)

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and should be used in future publications as defining factors for RS.

A second hurdle in relation to the diagnosis of RS is the quantified assessment of this triad. The diagnosis largely remains subjective and is often only evaluated by the visual judgment of a physician.

Up till now, there have been several attempts to measure micrognathia, and various methods have been proposed to objectively measure mandibular size. With computed tomography (CT) scans, it is possible to create 3-dimensional (3D) generated images of the mandible, which allows for precise measurements. However, radiation exposure during infancy and the lack of normative CT data are shortcomings that prevent routine adoption of CT scans for mandibular assessment. At the moment, CT scans are only performed in several countries to assess mandibular bone stock when mandibular distraction osteogenesis is considered a treatment option (Roy et al., 2009; Mahrous Mohamed et al., 2011; Chung et al., 2012; Meyers et al., 2015; Lopes et al., 2016; Tucunduva et al., 2016; Ramieri et al., 2017).

Micrognathia can also be assessed with plaster casts, 3D photogrammetry and direct measurements with rulers and calipers (Breugem et al., 2016). Unfortunately, no gold standard exists for the diagnosis of micrognathia since all these methods differ in accessibility, lack reliability studies, and act as poor methods to monitor mandibular growth.

Van der Haven et al. (1997) published a study on assessing micrognathia using a jaw index. They measured facial dimensions on 100 healthy newborns and 4 patients with RS aged younger than 1 week and defined the jaw index as “Alveolar overjet \times (Maxillary arch/Mandibular arch),” measured in millimeters. Mandibular anomalies are common in neonates, and micrognathia has been described in more than 100 syndromes (Evans et al., 2011). Both micrognathia and retrognathia involve abnormal, arrested development of the mandible. Although often difficult to distinguish, micrognathia refers to the size of the mandible, and retrognathia refers to the posterior mandibular positioning in relation to the maxilla. Both are relevant in the development of glossoptosis in neonates with RS, as the limited oral cavity size pushes the tongue base back in the pharyngeal space. The jaw index measures the maxillary–mandibular discrepancy by quantifying the micrognathic and retrognathic jaw by its alveolar relationship with the maxillary development.

The average jaw index in the study of Van der Haven and colleagues was 15.3 (± 1 standard deviation or SD) in the RS group compared to 4.2 (1.8SD) in the control group, implicating a 3.6 times higher value of the jaw index in patients with RS compared to healthy controls. Due to the small numbers of participating patients with RS, and the absence of studies on intra- and interobserver reproducibility, the reliability of this diagnostic tool for the assessment of mandibular proportions remains unknown.

Nevertheless, objective determination of the triad of RS is essential for adequate comparisons and harmonization of management strategies and eventually for the development of

future evidence-based treatment guidelines. So, during the Robin Sequence Consensus Meeting in 2014 in Utrecht, several recommendations were made in an attempt to more objectively assess this triad. Polysomnography (PSG) was recommended to establish UAO in children with suspected RS. To determine glossoptosis, a minimum of an endoscopy was recommended for the visualization of the tongue position in the oral cavity and oropharynx. To determine micrognathia/retrognathia, the jaw index was considered the best option available as a non-invasive tool in the workup of patients with RS.

The jaw index is noninvasive, fast, and easy to perform in almost any health-care situation, including those with no access to high-tech equipment (Breugem et al., 2016), but there is still lack of evidence about its reliability. Therefore, in this present study, the reliability of the jaw index in the neonatal mandible, measured with a specially developed caliper, is studied in further detail.

Materials and Methods

From February until April 2017, 52 healthy newborns were enrolled for inclusion at the obstetric and delivery ward at our hospital.

All newborns were included by one of the authors in collaboration with the obstetrician or nurse, and newborns were defined as healthy if they could be discharged within 1 week after birth.

Newborns were included if they were born full term (≥ 37 weeks gestational age), without any respiration or feeding problems and without congenital malformations or facial deformities due to birth trauma (eg, breech position). Both parents had to sign an informed consent. Patients were excluded if they were uncooperative or parents were unable to read and understand the written information.

For this study, the dimensions of the jaw index were done following the protocol of Van der Haven et al. (1997). The jaw index is defined as “Alveolar overjet \times (Maxillary arch/Mandibular arch),” measured in millimeters. For the measurements, 3 facial features were important. First the tragon, described as the point situated in the notch just above the tragus of the ear and which is visual as being a small projection in front of the external opening of the ear. Furthermore, the pogonion, described as the most projecting median point on the anterior surface of the chin. Finally, the subnasal point, described as the point where the nasal septum and the upper lip meet in the midsagittal plane. Every measurement was done with the neonate in a lying position. To measure the maxillary arch and the mandibular arch, no distinction was made between an awake, asleep, or a crying neonate. But for the alveolar overjet measurements, if the neonate was crying, we decided to come back at a later moment that day.

The mandibular arch was measured from the left tragon via the pogonion to the right tragon. The maxillary arch was measured from the left tragon via the subnasal to the right tragon. The alveolar overjet was assessed in a frontal-dorsal direction, between the most anterior points of the alveolar arches of the maxilla and the mandible (Figure 1).

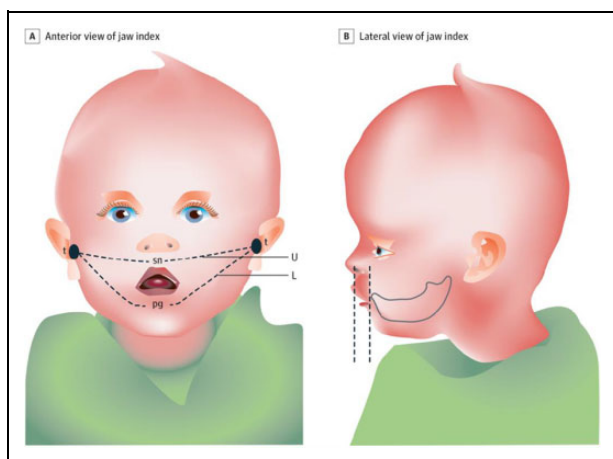


Figure 1. Jaw index. (Breugem et al., 2016)

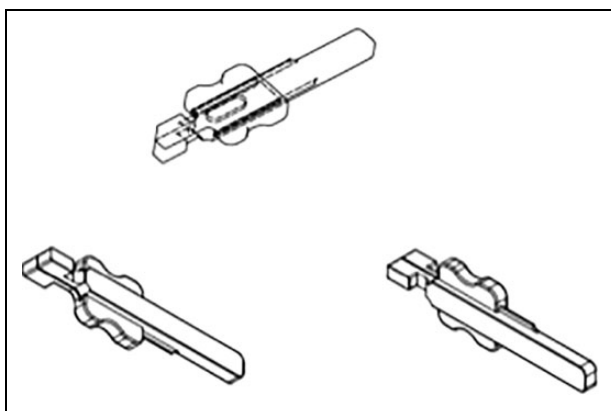


Figure 2. Alveolar overjet building plan—overview.

In the article of Van der Haven et al. (1997), a tire tread depth caliper was used for measuring the alveolar overjet. In this study, the alveolar overjet was measured with an instrument designed and fabricated by the physics and medical technology department of our hospital. This department also tested the safety of the instrument and provided a user manual. Currently, the instrument is not commercially available but could be easily fabricated by our or other technology department after its reliability has established. The building design has been added to this article Figure 2 and Supplemental files 1 and 2).

The first 2 authors of this article collected all data. Each child was measured 3 times. Beforehand, via block randomization with block size 4 and a 1:1 ratio, a statistician randomized which of the 2 authors performed a second measurement.

For all measurements, blinding precautions were taken into account. The mandibular and maxillary arches were assessed with a blank tapeline. For every measurement, 2 tapelines were needed (1 for the mandibular arch and 1 for the maxillary arch). For the second measurement of the same author, 2 new blank tapelines were used, implicating that we needed a total of 6 blank tapelines for every neonate. Each measurement was done by placing the end of each tapeline on the tragon of the

left ear, bringing it via the pogonion or subnasal point to the tragon of the right ear. Then, the distance was marked on the tapeline with a pen. After marking the 6 measurements, the exact distance on the blank tapeline was established in millimeters with a measuring lint.

For the alveolar overjet, our novel instrument was designed without a millimeter distribution for blinding reasons. As placing the millimeter distributor on the instrument could interfere with the intra-rater reliability. The results were measured after completion of all measurements with a measuring lint in millimeters.

After a thorough evaluation, the Medical Research Involving Human Subjects Act (WMO) declared that an official approval by their committee for this study would be unnecessary (WMO submission number: VUmc2015-596).

Statistical Analysis

We conducted an a priori power analysis to determine the required sample size. The power analysis was performed based on data from the earlier study of Van der Haven et al. (1997).

To detect an interclass correlation coefficient and intraclass correlation coefficient (ICC) of 0.8 with a 95% confidence interval (95% CI) of width 0.3, we needed 52 healthy patients when 2 observers measure the micrognathia (Shoukri et al., 2004). The interclass correlation coefficient is defined as the reliability of one measurement between observers. The ICC is defined as the reliability of 2 measurements by the same observer.

The data were analyzed using SPSS Statistics version 22 (IBM Corp, Armonk, New York). The interclass correlation coefficients for the jaw index were calculated with the 2-way random model and the ICC with the 2-way mixed model, where only the 2 measurements from the same observer were used. The ICC was also calculated using the covariance parameters of a linear mixed model, to link each second measurement to the right observer. In this model, we included a random effect for the participant and the observer as well as their 2-way interaction. However, 95% CIs could not be computed. Therefore, we chose to report the ICC via the 2-way mixed model above.

An ICC was classified as poor (<0.20), fair (0.21-0.40), moderate (0.41-0.60), good (0.61-0.80), or excellent (0.81-1.00).

Bland-Altman plots were used to assess repeatability of the measurements between the 2 different observers as well as between the 2 measurement of the same observer and limits of agreement ($LOA = \text{mean difference} \pm 1.96 \times \text{SD of the difference}$) were calculated.

Pearson correlation coefficients were computed to study the relation between age and weight with the jaw index, mandibular arch, maxillary arch, and alveolar overjet. A *P* value of less than .05 was classified as a significant correlation. For this, the average of the first measurement of each observer was used. Descriptive statistics for these individual parameters are presented as means and SDs.

Although not examined as a primary outcome for this study, we evaluated the possible existence of a learning curve in determining the jaw index. The group was divided into 3 groups of 17 or 18 healthy subsequently measured newborns. The interclass correlation coefficient and ICC were determined for each of these groups separately. For this assessment, no power analysis was performed. Therefore, this result is considered an explorative outcome.

Results

Population

Of the 52 included healthy children, 50% were male and 50% were female. The median age was 1.00 day (interquartile range [IQR] 1.00-1.75) and gestational age was 39.14 weeks (IQR 38.18-39.96). Most neonates were born by cesarean delivery (61.5%). Demographic data are summarized in Table 1.

Table 1. Demographic Data.

| | Median (25-75 Percentiles) |
|-------------------------|----------------------------|
| Age (days) | 1.00 (1.00-1.75) |
| Gestational age (weeks) | 39.14 (38.18-39.96) |
| Weight (g) | 3261 (2841-3753) |
| | N (%) |
| Vaginal delivery | 20 (38.5) |
| Cesarean delivery | 32 (61.5) |
| Gender | |
| • Female | 26 (50) |
| • Male | 26 (50) |

Interclass and Intraclass Correlation

The interclass correlation coefficient between the 2 observers was good with 0.74 (95% CI: 0.49-0.86), whereas the intraclass correlation coefficient between 2 measurements by the same observers was excellent with 0.81 (95% CI: 0.66-0.89; Table 2). The intraclass correlation coefficient based on the linear mixed model with random effects for the 2 observers was slightly higher (0.86), but no 95% CI could be computed.

Mean difference between measured jaw index of observers A and B was -0.31 (95% CI: -0.47 to 0.14), with LOA of -1.5 (95% CI: -1.8 to -1.2) to 0.88 (95% CI: 0.59 - 1.2).

Mean difference between measurements of the same observer was 0.005 (95% CI: -0.15 to 0.17) and the LOA -1.1 (95% CI: -1.42 to -0.87) to 1.2 (95% CI: 0.88 - 1.4 ; Figure 3).

There was a proportional error as variability was not correlated with the size of the jaw index.

To explore a possible learning curve for this instrument, we divided the group in 3 consecutive groups of 17, 18, and 17 newborns and compared the interclass correlation coefficient and ICC of the different groups.

In the first group, the interclass correlation coefficient was 0.59 (95% CI: -0.06 to 0.85), in the second group 0.82 (95% CI: 0.52 - 0.93), and in the third group 0.72 (95% CI: 0.16 - 0.90).

The ICC of the first group was 0.70 (95% CI: 0.15 - 0.89), second group 0.74 (95% CI: 0.30 - 0.90), and in the third group 0.89 (95% CI: 0.70 - 0.96).

The ICCs based on the linear mixed model with random effects for the 2 observers were lower: 0.46 for the first group, 0.69 for the second, and 0.61 for the third group.

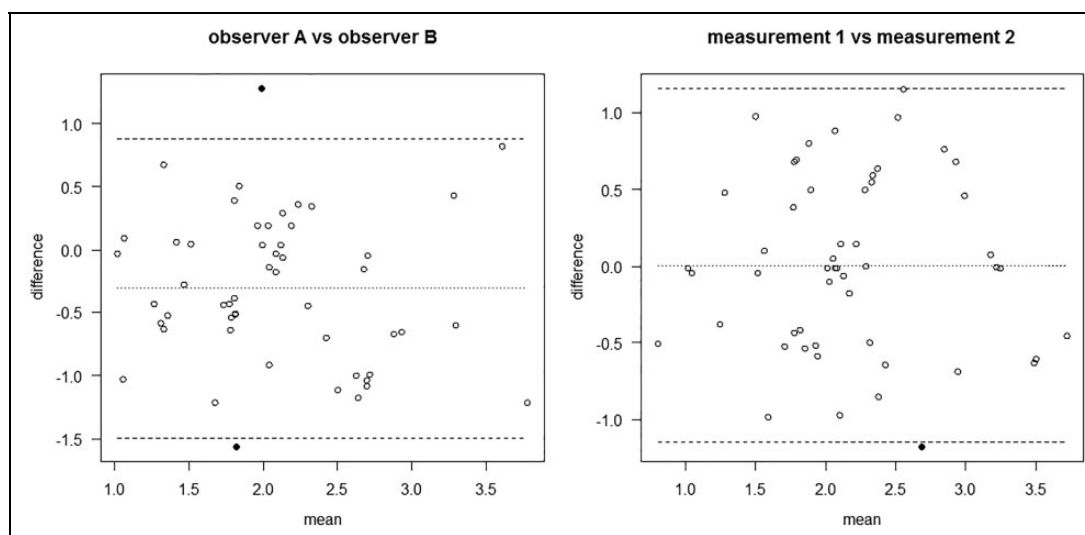


Figure 3. Bland-Altman plot for repeated measurements showing the comparison of the measured jaw index between observer A and B and between measurement 1 and 2 of the same observer. The mean between these two values and the upper and lower limits of agreement (LOA) are presented.

Table 2. Primary Parameters and Individual Parameters With Their Correlation With Age and Weight.

| Primary Parameters and Learning Curve | | | | |
|---|---------------------------------------|--------------------------|---------------------------|--------------------------|
| Primary parameters | | | | |
| Intraclass correlation | 0.81 (95% CI: 0.66-0.89) | | | |
| Interclass correlation | 0.74 (95% CI: 0.49-0.86) | | | |
| Learning curve | | | | |
| Interclass | Group 1: 0.59 (95% CI: -0.06 to 0.85) | | | |
| | Group 2: 0.82 (95% CI: 0.52-0.93) | | | |
| | Group 3: 0.72 (95% CI: 0.16-0.90) | | | |
| Intraclass | Group 1: 0.70 (95% CI: 0.15-0.89) | | | |
| | Group 2: 0.74 (95% CI: 0.30-0.90) | | | |
| | Group 3: 0.89 (95% CI: 0.70-0.96) | | | |
| | Group 1: 0.46 ^a | | | |
| | Group 2: 0.70 ^a | | | |
| | Group 3: 0.61 ^a | | | |
| Individual Parameters and Correlation With Age and Weight | | | | |
| Individual parameters | | | | |
| | Maxillary Arch (mm) | Mandibular Arch (mm) | Alveolar Overjet (mm) | Jaw Index |
| | 168.7 (SD 9.4) | 162.6 (SD 11.1) | 2 (SD 0.6) | 2.0 (SD 0.64) |
| Correlation | | | | |
| Age | $r = 0.10$ $P = .46$ | $r = 0.051$ $P = .72$ | $r = -0.12$ $P = .41$ | $r = -0.11$ $P = .46$ |
| Weight | $r = 0.73$ $P < .001$ | $r = 0.71$ $P < .001$ | $r = -0.062$ $P = .66$ | $r = 0.086$ $P = .54$ |

Abbreviations: CI, confidence interval; SD, standard deviation.

^aCovariance parameters used to prevent underestimation or overestimation.

Individual Parameters

Descriptive statistics of each parameter are depicted in Table 2. None of the parameters correlated with age. The mandibular and maxillary arch correlated highly and significantly with weight (both $r > 0.7$, both $P < .001$), whereas alveolar overjet and the jaw index itself were not correlated with weight ($P \geq .54$; Table 2).

Discussion

Micrognathia has been defined as 1 of the 3 base characteristics of RS. Aside from being easy, simple, and noninvasive, the jaw index could be a useful diagnostic tool for objectifying micrognathia. Van der Haven et al. (1997) first described the jaw index and also claimed that the jaw index would increase with micrognathia and decrease whenever prognathia was present. An omission in their study was that the reliability of this diagnostic tool was not tested. Consequently, the aim of our study is to assess the reliability of the jaw index in healthy newborns in order to compare these results with children clinically diagnosed with RS. This would allow us in the future to use this instrument as a diagnostic tool for micrognathia in suspected RS cases.

The results of this study demonstrate a good interclass correlation coefficient of 0.74 and an excellent intraclass correlation coefficient of 0.86, demonstrating that when 2 different individuals assess the same child, the jaw index is a consistent tool between observers. When one person assesses a child twice, we see an excellent reproducibility, indicating that the

measurements are also consistent within one individual observer.

Besides the study of Van der Haven and colleagues (1997), no literature is available concerning the reliability of the jaw index or the average length of the different jaw index parameters assessed on newborn children. Only in the study of Paladini et al was the jaw index studied in utero with 2D and 3D ultrasound from 10 weeks of gestation (Paladini et al., 1999). As a different measurement method was used and all these children were measured in utero, no comparison can be made with our results.

With regard to the length of the mandibular arch, maxillary arch, and the alveolar overjet in newborn children, Van der Haven et al. (1997) revealed a mean maxillary arch of 168 mm (range 139-194 mm), a mean mandibular arch of 159 mm (range 131-188 mm), a mean alveolar overjet of 4.0 mm (range 0.5-9.0 mm), and a mean jaw index of 4.2 (range 2.4-6). Comparing these results with our study, a different alveolar overjet (4.0 mm vs 2.0 mm) and consequently a different jaw index (4.2 vs 2.1) were demonstrated. Two important confounders could explain the difference between these results. First, the newborn children measured in our study had a median age of 1 day. The study of Van der Haven et al. (1997) did not reveal the exact age of the children during their measurements, but all the children were younger than 1 week. Thus, there is a possibility that the children in their study were older than 1 day, which could influence the size of the jaw (Vegter et al., 1999).

Second, and probably the most important confounder was the use of a different measurement instrument in our study for the assessment of the alveolar overjet. In the study of Van der Haven et al. (1997), a tire tread depth caliper was used instead of a specially designed instrument, as was done in our study.

To give an estimation of the jaw index assessed on patients clinically diagnosed with RS, we compared our results with a retrospective study performed at our center. In this study, the jaw index was measured on 28 patients with RS treated with a tongue lip adhesion between 1993 and 2016 (Mermans et al., 2018). The mean jaw index was 13.2 measured according to the Van der Haven protocol (1997). This is a 6.3 times higher index compared to the jaw index of 2.1 measured on the 52 healthy newborns in this study. Unfortunately, these findings are non-comparable, giving the methodological difference among these studies.

To consider the reproducibility and user friendliness of the diagnostic tool, we analyzed the required learning curve for this instrument. We found an interclass correlation coefficient in the first 17 newborns of 0.59 (95% CI: -0.06 to 0.85), in the second 18 newborns of 0.82 (95% CI: 0.52-0.93), and the last 17 newborns of 0.72 (95% CI: 0.16-0.90). The corrected intraclass correlation coefficients in these 3 groups were, respectively, 0.46, 0.69, and 0.61. This means that this tool seems to have a learning curve in at least the first 17 patients. However, the aim of this study was not to calculate the learning curve of this tool, so the abovementioned claim is statistically underpowered due to the small sample size. Consequently, the results have to be carefully interpreted.

Nevertheless, throughout the measurements, we did notice a limitation while measuring the alveolar overjet. Therefore, some specific elements have to be taken into account. First, it is better to do the measurement after the baby has eaten, as this minimizes the risk of crying and avoids the need to come back later to do the measurements. Second, all equipment must be prepared in advance to perform these measurements promptly, since these young neonates get irritated easily and as a consequence could start crying. A crying neonate makes the alveolar overjet instrument impossible to handle as the child opens its mouth in such an extent that the lower guidance does not contact the mandibular dental alveolar bone or the upper guidance does not contact the maxillary dental alveolar bone.

Furthermore, it is unknown how relaxation and tightening of the mandible of a crying neonate compared to a resting neonate affects the reliability of the measurement of the overjet size. With this study, we cannot provide this information. Further research on the influence of mandibular relaxation and tightening on the overjet would be interesting.

For future measurements and until there is evidence about the influence of the jaw position on the overjet size, it is imperative to maintain measurements as consistent with the measurements done in this study.

In patients with breathing problems with suspected RS, we would advise to take the measurements with the child in a supine position. This would be ideally performed during a PSG or before surgery. We would like to stress that the patient

should not be intubated or have had muscle relaxants administered, yet as these could influence the position of the jaw.

Catch-up growth of an underdeveloped mandible is another important topic in the literature. Many studies have dedicated their research on the measurement of catch-up growth of the mandible in children believed to have RS. Some investigators have supported the compression theory, according to which micrognathia of the mandible is the result of intrauterine molding against the sternum (Ozawa et al., 2012). If this theory is accurate, it would appear logical to expect some rebound growth of the mandible shortly after birth, reducing facial convexity and perhaps allowing the mandible to catch up with the maxilla (Figueroa et al., 1991; Ozawa et al., 2012). All studies not supporting the theory of mandibular catch-up growth are unanimous in describing that the mandible remained micrognathia in RS (Vegter et al., 1999; Hermann et al., 2003; Suri et al., 2010; Ozawa et al., 2012). By measuring the jaw index of children with RS at several standardized intervals (eg, every 3 months until the age of 1 year), more research can be done on which children show catch-up growth, without the need for several CT scans.

Moreover, for future research, the jaw index could be compared in patients who also had a CT scan to correlate the findings between these research modalities. In addition, the jaw index could be compared in patients who had a PSG to determine whether there is a correlation between the mandibular size and degree of UAO.

In this study, we reported the reliability of the jaw index presented as the interclass correlation coefficient and the intraclass correlation coefficient, and the normal values of the jaw index in healthy newborns. The validity of this instrument is not tested. Therefore, for future research, construct validity should be tested on patient clinically suspected of RS in order to verify our instrument truly measures micrognathia.

As the validation of an instrument is a continuous process in which refinement of a theory strengthen the validation of the measurement instrument, for future research, this instrument should also be validated for the different possible jaw positions on healthy newborns (De vet et al., 2011).

These findings could be clinically relevant to physicians, as the jaw index is a simple and noninvasive tool that can be used to diagnose micrognathia in children suspected of having RS without the need for exposure to radiation.

Conclusion

The jaw index is a reliable instrument used to objectify the size of the lower jaw compared to the upper jaw in healthy newborns, both for the intraobserver and interobserver reproducibilities. The reliability and validity of this instrument for the assessment of micrognathia in children with suspected RS seems likely. Nevertheless, in order to verify the diagnostic added value of this instrument for micrognathia, further research regarding this is needed. Testing the construct validity of this instrument would be required through refinement of our hypotheses on healthy newborns to be able to validate

this instrument on patients clinically diagnosed with RS in the future.


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Supplemental Material

Supplemental material for this article is available online.

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