

Clinical and Fiberoptic Endoscopic Evaluation of Swallowing in Robin Sequence Treated With Nasopharyngeal Intubation: The Importance of Feeding Facilitating Techniques

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Objective: To evaluate oral feeding capacity, the swallowing process, and risk for aspiration, both clinically and during fiberoptic endoscopic evaluation of swallowing, in infants with isolated Robin sequence treated exclusively with nasopharyngeal intubation and feeding facilitating techniques.

Design: Longitudinal and prospective study.

Setting: Hospital de Reabilitação de Anomalias Craniofaciais, University of São Paulo, Bauru, Brazil.

Patients: Eleven infants with isolated Robin sequence, under 2 months of age, treated with nasopharyngeal intubation.

Interventions: Feeding facilitating techniques were applied in all infants throughout the study period. The infants were evaluated clinically and through fiberoptic endoscopic evaluation of swallowing at first, second, and, if necessary, third week of hospitalization (T1, T2, T3). The mean volume of ingested milk was registered during clinical evaluation, and events were registered during feeding.

Results: The respiratory status of all infants was improved after nasopharyngeal intubation; 72% of them presented risk for aspiration during fiberoptic endoscopic evaluation of swallowing at T1. This risk was less frequent when thickened milk was given to the infants and at subsequent evaluations (T2 and T3).

Conclusions: Nasopharyngeal intubation aids in stabilizing the airway in isolated Robin sequence, but it does not relate directly to feeding. The risk for aspiration was present in most of the infants, mainly during the first week of hospitalization, and improved within a few weeks, after the use of feeding facilitating techniques.

KEY WORDS: *airway obstruction, dysphagia, Pierre Robin sequence, swallowing disorders*

Robin sequence (RS), defined as micrognathia and glossoptosis, with or without cleft palate, is characterized clinically by obstruction of the upper airway and by respiratory and feeding difficulties, which are more frequent and more severe during the first months of life. The respiratory difficulty is aggravated during sleep. This

anomaly may arise as an isolated entity called isolated Robin sequence (IRS), as a component of a known syndrome, or in association with other malformations that do not characterize a known syndrome (Shprintzen, 1992; Marques et al., 1998). Prolonged nasopharyngeal intubation (NPI) has been used as a treatment modality even in patients with severe respiratory obstruction (Heaf et al., 1982; Marques et al., 2001b; Wagner et al., 2003; Marques et al., 2005; Marques et al., 2008). The efficacy of NPI in improving the respiratory difficulties of small infants with RS has been demonstrated by different investigators (Heaf et al., 1982; Marques et al., 2001b; Wagner et al., 2003; Anderson et al., 2007).

Although different mechanisms of upper airway obstruction have been classified into four types (1, 2, 3, and 4) by Sher et al. (1992) and Sousa et al. (2003), the most frequent mechanism in IRS is type 1 (i.e., the airway obstruction resulting from posterior displacement of the tongue and dorsum touching the posterior wall of the pharynx). The

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other types, mainly 3 and 4, are much more frequent in RS associated with syndromes and involve dysfunction of airway muscles.

The oropharyngeal dysphagia in IRS results mainly from upper airway obstruction and tongue retroposition. This is aggravated by cleft palate, but there is no evidence that the cleft palate itself places the infant at risk for aspiration (Masarei et al., 2007). A study performed in the Hospital de Reabilitação de Anomalias Craniofaciais–University of São Paulo (HRAC-USP) demonstrated that oral feeding capacity in infants with RS treated with NPI improved after the introduction of some techniques specially developed for these infants, denoted as feeding facilitating techniques (FFT) (Nassar et al., 2006).

The NPI and FFT have permitted 85% of infants with IRS to receive hospital discharge in fewer than 15 days, with exclusively oral feeding (Nassar et al., 2006).

The experiences at the HRAC-USP with infants treated with NPI indicate immediate improvement of respiratory difficulty; whereas, several days are necessary for improvement of oral feeding difficulty, despite the use of FFT (Marques et al., 2008).

Some questions about the influence of NPI in the swallowing process have arisen: (1) Will infants treated with this procedure have an aspiration risk during oral feeding? (2) Why does the feeding difficulty remain after the upper airway obstruction is released? The first question is the purpose of this study.

Fiberoptic endoscopic evaluation of swallowing (FEES) is a simple and safe method that evaluates swallowing and provides information about patients with oropharyngeal dysphagia (Langmore et al., 1988, 1991; Hartnick et al., 2000; Macedo Filho, 2000). Some authors (Langmore and MacCulloch, 1997; Willging, 1997) have stated that this exam can be performed in small infants and is well tolerated by them. It offers information about the anatomy and function of some of the structures involved in the deglutition process. This exam yields useful information for the diagnosis and decisions regarding treatment and evaluates aspiration risk during feeding in infants with swallowing problems. The FEES does not submit the child to radiation, which permits the exam to be performed frequently.

The objectives of the present study were (1) to evaluate, clinically and through FEES, oral feeding capacity, the swallowing process, and risk for milk aspiration in infants with RS treated exclusively with NPI and FFT during the first weeks of treatment; (2) to evaluate the importance of FEES and FFT to the introduction of safe oral feeding in these infants.

METHODS

Eleven infants with IRS under 2 months of age admitted to HRAC-USP and in need of NPI as the only treatment modality for airway obstruction were evaluated. Preterm



FIGURE 1 Infant with nasopharyngeal intubation.

infants or infants with other diseases associated with IRS that could impact their physical condition were excluded from the study.

Nasopharyngeal intubation was performed when infants presented recurrent crises of pallor and/or cyanosis and/or apnea, O_2 saturation $<90\%$ measured by continuous pulse oxymetry. These infants frequently present with severe feeding difficulties and need feeding tubes. This procedure was performed during the first days of hospitalization and consisted of a 3- or 3.5-mm-diameter silicone tube, usually used for tracheal intubation. This tube was adapted to NPI as follows: cut at level 9 cm, introduced 7 to 8 cm through the nostril, fixed with surgical tape, cut 1 cm outside the nostril, and kept positioned slightly above the epiglottis (Marques et al., 2001a; Marques et al., 2005) (Fig. 1). The position of the NPI was confirmed by nasopharyngoscopy.

Improvement of respiratory distress with NPI was considered to have occurred when O_2 saturation, measured by continuous pulse oxymetry, was maintained above 90% in ambient air; when respiratory effort was reduced (reduction of pallor and cyanosis crises, of intercostal and supraclavicular retraction, and of inspiratory noise characteristic of glossoptosis observed by pulmonary auscultation); when it was possible to stimulate oral feeding, and

the child became comfortable with NPI, without accumulation of secretions and saliva in the oral and/or nasal cavity and/or in the tube for NPI.

The infants initially received their diet through a nasogastric tube, and oral feeding was gradually introduced according to the individual ability of the infant.

The infants were evaluated clinically and through FEES at three times: T1, T2, and, if necessary (i.e., if the child still presented any abnormal event during FEES at T2) at T3, which corresponds to the first, second, and third week of hospitalization, respectively.

Clinical evaluation consisted in determining the mean ingested milk volume for 30 minutes during oral feeding for three consecutive feedings and in registering events during oral feeding (i.e., vomiting, coughing, cyanosis, and decreasing oxygen saturation as measured by pulse oximetry). If the child presented one or more events, oral feeding was discontinued and the volume accepted was recorded. The oral feeding capacity was evaluated with and without NPI at T1, T2, and, if necessary, at T3, and the results were compared.

The FEES was carried out with an ENF-P4 Olympus flexible optic fiber with 3.4-mm external diameter (http://www.olympus-europa.com/endoecopy/427_ENF-P4.htm), an Olympus light source (http://www.olympus-europa.com/endoecopy/431_VISERA_CLV-S40.htm), and a Sony monitor (<http://www.sony.com.br/corporate?page=subcat&id=1488>). Examination data were saved on a videocassette for review and archiving. The exams were performed when infants were hungry and on the mother's lap so that they became fully cooperative, that is, calm and not crying, when the endoscope was in place. Anesthesia was not used in order to avoid reducing pharynx sensibility. First, the flexible fiberoptic scope was introduced into the nostril and, later on, advanced through nasopharynx and oropharynx. To observe swallowing, the flexible fiberoptic scope was positioned between the nasopharynx and the oral pharynx, and the milk was given through the oral route. Aniline color was added to the milk to make it blue, to contrast with the pink color of the oral mucosa and to facilitate observation of the milk during swallowing. Throughout the exam, milk was given through bottle and latex nipple, with different hole sizes: original, intermediate (larger than the original but smaller than 1 mm), and 1 mm. Milk was given in different consistencies: liquid or thickened liquid. The milk was thickened with a modified corn-based flour with a concentration of approximately 3% (i.e., thicker but still a liquid) (Fig. 2). Swallowing was evaluated with three different hole sizes, with fluid milk, and later on with thickened milk, with and without NPI. We considered as aspiration risk the following results of FEES: milk reflux into NPI and/or delayed initiation of swallowing (milk into the larynx before the "white out" that occurs as the pharyngeal swallow is initiated) and/or presence of milk residue on epiglottis and/or on vocal folds and/or in trachea (below vocal cords) after swallowing

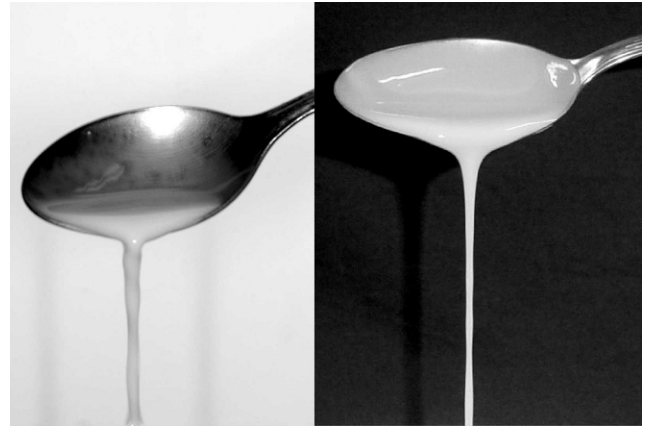


FIGURE 2 Liquid and thickened liquid.

when the epiglottis has returned to the rest position (Macedo Filho, 2000).

Weight gains, from admission to hospital discharge, were studied also. The birth weight measurements were obtained from information provided by the parents.

Criteria to determine capability for exclusive oral feeding included (1) ability to ingest 70% of milk volume recommended for the age, in fewer than 30 minutes, (2) no choking, and/or (3) no risk for aspiration observed by FEES. Other data recorded from each infant included (1) volume ingested orally per feeding at T1 and at T2 or T3, (2) number of days needed to reach totally oral feeding, and (3) number of days in hospital.

The influence of NPI in deglutition was determined clinically and through FEES from the first week of admission until hospital discharge, in a longitudinal and prospective study. The necessity for maintenance of this procedure at discharge was registered also.

The FFT were applied immediately after the first clinical evaluation and maintained during the whole study period. These techniques were used according to the necessity of the infants and consisted of the following: the use of a pacifier and massage in order to relax and move the tongue forward before each feeding if the infant presented severe retroposition of the tongue; manual support in order to maintain mandibular stability during feeding if the infant presented severe micrognathia; a long and soft nipple with a hole enlarged to 1 mm in diameter; insertion of the nipple exactly on the tongue during suction; and rhythmic movement of the nipple in the mouth during suckling if the infant presented severe sucking difficulty. These techniques are illustrated with photographs in a previous study performed at the HRAC-USP (Nassar et al., 2006). In the present study, it was necessary to introduce two new FFT: (1) manual support of the cheek to facilitate lip closure and to improve the effectiveness of sealing, avoiding oral milk escape (Fig. 3) and (2) thickening of milk to get a thickened consistency and reduce the reflux of milk into the NPI tube (Fig. 2). The FFT most used during the study were also registered.



FIGURE 3 The manual cheek support during nursing in an infant with Robin sequence and nasopharyngeal intubation.

The study was approved by the Research Ethics Committee of HRAC-USP (process n° 095/2004-SVA-PEPE-CEP) and written consent for the infants' participation in the study was given by all caretakers.

Statistical Analysis

The analysis of swallowing was based on the description of the results of clinical and FEES evaluations.

The Wilcoxon test (Hollander and Wolfe, 1999) was used to compare the median of the ingested mean volumes per feeding, at admission and at hospital discharge, and the median of the weight gain before and during hospitalization.

RESULTS

The mean age of the studied infants at admission to the HRAC-USP was 35 ± 18 days (Table 1). All infants had a cleft palate.

During the first week of the study, at T1 (5.81 ± 1.87 days of hospitalization), all 11 infants improved their

respiratory status with NPI, according to previous criteria, and needed it to maintain the patency of the airway. At T2/T3 (17.54 ± 3.44 days of hospitalization) and at hospital discharge, 10 infants (90%) needed this procedure to improve respiratory discomfort, and the one remaining maintained a good respiratory and feeding pattern without NPI (Table 2).

At T1, all infants showed improvement in the oral feeding capacity with NPI because respiratory distress improved with this procedure. At T2, T3, and at discharge, seven infants could be fed better orally with NPI than without it, but three infants had to have this procedure interrupted for feeding (Table 2). These three infants needed prolonged use of NPI to improve respiratory distress, mainly during deep sleep, but it was necessary to remove it during feeding due to increased risk for aspiration (i.e., the reflux of milk into the NPI tube). These infants did not present this risk during oral feeding without NPI. The prolonged use of NPI enlarged the nostril, making it easy to reintroduce the NPI tube, and the infants tolerated repetitive removal and reinsertion of the tube.

Of the 11 studied infants, seven (63.63%) presented with aspiration risk with NPI on first FEES at T1 (Table 3). In these infants the milk refluxed into the NPI tube during swallowing and returned into the airways during inspiration, which predisposed the infants to aspiration. These FEES events were reduced with thickened milk. These seven infants also presented with aspiration risk on first FEES without NPI (milk residue on vocal folds after the swallowing).

Of the seven infants who presented aspiration risk at T1, three did not present this risk at T2, with or without NPI; three presented this risk at T2 and T3 with NPI only; and one presented with aspiration risk on FEES at T2 and T3 with or without NPI (milk residue on vocal folds after the swallow) (Table 3).

Four infants (36.36%) did not present with abnormal findings on first FEES (T1) (Table 3), with or without NPI, and oral feeding could be introduced during the first week of hospitalization.

TABLE 1 Weight Values in Grams at Birth, at Hospital Admission, and at Hospital Discharge, Ages in Days at Hospital Admission and Discharge and Length of Hospitalization

Infant	Birth Weight (g)	Weight at Hospital Admission (g)	Age at Hospital Admission (d)	Weight at Hospital Discharge (g)	Age at Hospital Discharge (d)	Length of Hospitalization (d)
1	2620	2850	24	3750	52	28
2	2000	2280	42	3254	93	51
3	4000	3390	55	3765	78	23
4	3470	3080	13	3820	36	23
5	3390	3060	35	3370	52	17
6	3220	2900	52	3606	74	22
7	2405	2610	11	2760	26	15
8	3225	3320	47	3448	69	22
9	3250	2650	26	3130	46	20
10	2800	4200	63	4738	76	13
11	3000	2700	15	3140	25	10
Mean \pm SD	3034 \pm 553	3003 \pm 511	35 \pm 18	3525 \pm 516	57 \pm 23	22 \pm 11

TABLE 2 Distribution of Infants With Robin Sequence, Treated With NPI and FFT, in Accord With Feeding and Respiratory Behavior, at T1 and at T2/T3*

Infants' Behavior	T1 Number of Infants	T2/T3 Number of Infants
NPI was important to improve breathing and feeding.	11	7
NPI was important to improve breathing, but it was necessary to be removed during feeding.	0	3
NPI was not necessary for breathing or feeding.	0	1
The feeding was given exclusively by gavage.	7	1
The feeding was given by oral route, but it was necessary to be completed by gavage.	4	1
The feeding was given exclusively by oral route.	0	9

* NPI = nasopharyngeal intubation; FFT = feeding facilitating techniques; T1 = first week of hospitalization; T2 = second week of hospitalization; T3 = third week of hospitalization.

Of the seven infants who were discharged from hospital with prolonged use of NPI to improve respiratory and feeding difficulties, five (71.43%) presented with aspiration risk on FEES at T1. The mean time for disappearance of abnormal videoendoscopic findings in these infants was 10.80 ± 9.65 days after the beginning of oral feeding stimulation with FFT.

All 11 studied infants needed feeding tubes for feeding at T1, seven (63%) needed feeding tubes only to complete their feeding, and nine (81.81%) acquired exclusive oral feeding capacity at T2/T3 and at hospital discharge (Table 2). The median volume of ingested milk at T1 was inferior to the median volume at T2/T3 ($p = .001$) (Table 4).

The FFT most used were as follows: use of pacifier (11 cases); increase of nipple hole (nine cases); thickening of milk (nine cases) (Fig. 2); massage to relax and anteriorize the tongue (11 cases); insertion of the nipple exactly on the tongue (11 cases); rhythmic movements of the nipple in the mouth (six cases); manual support of the cheek to improve the lip closure (three cases) (Fig. 3); and manual support of the mandible (one case).

The duration of hospitalization was 22 ± 11 days (Table 1).

The studied infants presented statistically significant weight gain ($p = .001$) during the period of hospitalization in comparison to the weight gain from birth to admission to the hospital (Tables 1 and 5).

TABLE 3 Aspiration Risk Observed on FEES in Infants With Isolated Robin Sequence Treated With NPI, During the First, Second, and Third Weeks of Hospitalization, Prospectively

Number of Infants	T1 (1st Week)	T2 (2nd Week)	T3 (3rd Week)
With aspiration risk on FEES	7	4	4*
Without aspiration risk on FEES	4	7	(7)†
Total	11	11	4

* Three of these infants did not present risk for aspiration without nasopharyngeal intubation (NPI) during oral feeding, observed on fiberoptic endoscopic evaluation of swallowing (FEES), and one presented aspiration risk with or without NPI.

† In seven infants the third FEES was not performed because they did not have risk for aspiration observed on FEES at T2.

TABLE 4 The Mean Volume of Orally Ingested Milk (mL), per Feeding, of Infants With Robin Sequence, at T1 (5.81 ± 1.87 Days) and at T2/T3 (17.54 ± 3.44 Days)†

Infant	T1 Mean Volume (mL)	T2/T3 Mean Volume (mL)*
1	0	60
2	25	20
3	60	60
4	15	40
5	20	50
6	20	60
7	47	60
8	35	45
9	40	50
10	37	60
11	15	45
Median	25	50

† T1 = first week of hospitalization; T2 = second week of hospitalization; T3 = third week of hospitalization.

* $p = .001$.

DISCUSSION AND CONCLUSIONS

In the literature, *oropharyngeal dysphagia* is defined as any abnormality in the swallowing physiology of the upper aerodigestive tract (Martino et al., 2000). It consists of a swallowing difficulty that may or may not compromise nutritional status and pulmonary function and is considered a symptom of an illness with aspiration risk (Furkim et al., 1999; Arvedson, 2005).

The instrumental examination of swallowing was needed to define the pharyngeal physiology. The FEES was useful for identifying some aspects of the pharyngeal phase of swallowing and for providing information that aided the decision regarding oral feeding management. Despite the invasive nature of this exam, it is well tolerated by very young children.

NPI has been used at HRAC-USP on a large-scale basis for the treatment of severe cases of RS (Marques et al., 2001a; Marques et al., 2001b; Marques et al., 2004). The ideal procedure would be to start the treatment during the very first days of life; however, because HRAC-USP is one of the major referral centers for craniofacial malformations in Brazil and receives patients from distant locations, this is

TABLE 5 Weight Gain (g/d) From Birth to Hospital Admission and From Hospital Admission to Hospital Discharge

Infant	Weight Change (g/d), Birth to Admission	Weight Change (g/d), Admission to Discharge*
1	+9.58	+32.00
2	+6.60	+19.10
3	-11.09	+16.30
4	-30.00	+32.20
5	-9.43	+8.90
6	-6.15	+32.10
7	+18.60	+10.00
8	+2.00	+5.80
9	-23.07	+24.00
10	+22.20	+41.40
11	-20.00	+44.00
Median	-6.00	+24.00

* $p = .001$.

not always possible. Another limitation of this study is that it was a small series with no control groups. The IRS is a rare malformation, and invasive exams in a control group would not be approved by the Research Ethics Committee.

There are no studies in the literature on RS that evaluate the influence of FFT in reducing aspiration risk or that use FEES to evaluate the safety of oral feeding. A previous study, performed in our institution (Nassar et al., 2006), demonstrated that FFT fostered oral feeding capacity in infants with RS but did not show the cheek support and the thickening of milk as FFT in RS. The cheek support is performed by pressing the cheek near the mouth (Fig. 3), and it was introduced in the present study to improve lip closure and to prevent oral escape of milk.

Pinelli and Symington (2005) showed strong evidence for using pacifiers to improve oral feeding in preterm infants, and Sheppard and Fletcher (2007) considered as weak the evidence for motor therapy in neonates. There are no studies in the literature with evidence that separate techniques such as the use of a pacifier, tongue massage, and rhythmic movement of the nipple could be efficient in IRS. However, Nassar et al. (2006) demonstrated that these techniques used together were able to improve oral feeding capacity in infants with IRS within 7 days.

Changing milk to a thickened liquid (Fig. 2) also was introduced in the present study as a new FFT for IRS, based on the pre-established concept that proposes a change in the texture of the milk to improve coordination and safety of deglutition (Arvedson and Brodsky, 2002). This technique reduced the aspiration risk, mainly in infants with IRS who presented reflux of milk to the interior of NPI tube during deglutition. The thickening of the milk reduced this reflux and, consequently, the return of refluxed milk to the larynx during inspiration.

The presence of cleft palate reduces the infant's ability to build up sufficient intraoral negative pressure for effective sucking to extract milk from the nipple (Mizuno et al., 2002; Masarei et al., 2007; Reid et al., 2007). The increased hole size in the nipple facilitates the infant's ability to extract milk (Nassar et al., 2006). However, the infant may work hard to slow the flow and have more difficulty if the liquid is extracted too fast with too much volume, which in turn may increase the risk for aspiration, likely before initiation of a pharyngeal swallow (Arvedson and Brodsky, 2002). These infants often cannot coordinate sufficiently the sequencing of sucking, swallowing, and breathing to take nipple feeds safely. A slow-flow liquid may give them more time to suck and coordinate swallowing (Arvedson and Brodsky, 2002). In the present study, FEES was important as one of the bases for optimal decision making related to consistency of the liquid to be given to infants with IRS.

We observed, in some of the patients, the need to remove the NPI during oral feeding due to the risk of aspiration and also the need to maintain this procedure beyond feeding time to improve breathing, mainly during deep sleep.

All the techniques used in this study let most of the infants be discharged from the hospital with exclusively oral feeding. The mean volume of ingested milk, in a period of less than 30 minutes, was significantly higher at discharge than at admission to the hospital, suggesting improvement of the dysphagia. The mean length of hospitalization was shorter than 30 days (Table 1), similar to other studies of infants treated with NPI (Marques et al., 2001b; Nassar et al., 2006; Anderson et al., 2007).

The mean age of the studied infants at admission to the HRAC-USP was 34 ± 18.24 days (Table 1); only three infants were 15 days or younger. Eight were 23 days or older and were expected to have gained weight, but instead most of them lost weight (median, -6 g) before admission to the hospital. Most infants (91%) presented recovery of nutritional status after hospitalization.

In conclusion, NPI aids in stabilizing the airway in infants with IRS, but it does not relate directly to feeding. Different FFT had to be applied to these infants because there was no single infant feeding behavior. Despite this heterogeneity, aspiration risk was present in most of the infants, primarily in the first week of hospitalization. The improvement occurred after a few weeks with the use of FFT. The frequency in the use of these techniques also was heterogeneous. Therefore, each case had to be considered individually and studied over time. The FEES was useful to verify aspiration risk and to indicate how to safely feed infants with RS treated with NPI. This exam plus clinical evaluation, performed sequentially, gave support to indicate the most appropriate time to start oral feeding and indicated the techniques to improve it.

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