### Nonnutritive sucking and upper airway instability

Suzione nonnutritiva ed instabilità delle prime vie aeree

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#### Introduction

In infants with an upper airway instability, the obstruction of the pharynx due to aspiration of the tongue (called «vacuum-glossoptosis apnoea») may play a role into the pathogenesis of sudden infant death syndrome (SIDS) 1-3. As non-nutritive sucking can stabilize the upper airway 4, in 1979 Cozzi et al. speculated that the use of a dummy may protect from glossoptosis-related SIDS 2. A recent study has been designed to test this hypothesis. Actually, Mitchell et al. have found that in a group of 485 SIDS victims the use of a dummy during the last sleep before death was very much less frequent than in a large control group of infants 5. They have calculated that by encouraging dummy use the number of SIDS victims in New Zealand could drop by about 50 percent 5. This epidemiological study supports the concept that SIDS may be related to glossoptosis-apnoea and reproposes the question: how can the use of a dummy stabilize the upper airway?

To study the possible mechanisms involved in this action, we review the mechanisms of various methods that have been found valid in clinical practice for the prevention of glossoptosis-apnoea associated with congenital micrognathia, hypertrophic adenoids, upper respiratory tract infection (URTI), or choanal atresia. These clinical observations are relevant because the pathogenesis of glossoptosis-apnoea associated with these conditions is similar to that postulated for glossoptosis-related SIDS <sup>1-3</sup>.

Methods which may prevent glossoptosis-apnoea in infants and children with congenital micrognathia and hypertrophic adenoids

Open-mouth breathing. In 1923, Pierre Robin coined the

terms «glossoptosis», meaning obstruction of the pharynx due to a relapse of the tongue, and «glossoptosism», meaning upper airway instability 6. He differentiated between a «congenital» and an «acquired» glossoptosis, the latter being more often associated with hypertrophic adenoids. In his original view, glossoptosis was a mechanical consequence of a congenital micrognathia 6. Pierre Robin noticed that the micrognathic infant breathes with his or her mouth open as closing his or her mouth causes a glossoptotic pharyngeal obstruction 67. Oral breathing, however, predisposes to URTI which in turn causes hypertrophy of adenoids and obstruction of the rhinopharynx 7. Actually the removal of the nasal obstruction by adenoidectomy does not cure «glossoptosism». This status is responsible for recurrent functional obstruction of the pharynx and persistent mouth-breathing that often continue after surgery («adenoidism without adenoids») 67. Open-mouth-breathing, therefore, does not appear to be the result of either a nasal obstruction or of a bad habit, but a manoeuvre adopted by the glossoptotic patient to bring the tongue forward, thus preventing glossoptotic pharyngeal obstruction 67. Our clinical experience supports the validity of this Robin's old and rather neglected concept.

Pierre Robin considered thumb sucking with an open mouth a pathognomonic sign of an upper airway instability <sup>7</sup>. A similar sign known as «the two fingers sign» has been more recently described in a group of young children with hypertrophic adenoids and sleep-apnoca <sup>8</sup>. The child may learn to prevent or to reverse severe glossoptosis-apnoea associated with hypertrophic adenoids by depressing his or her tongue with two fingers <sup>8</sup>. We, too, have noticed that the micrognathic infant may use the thumb to depress his or her tongue, thereby relieving respiratory distress and/or oropharyngeal dysphagia. Depressing the tongue is an action that involves passive opening of the mouth.

The pharyngeal vacuum. In infants with congenital micrognathia, measurements of pharyngeal pressure (Fig. 1) have shown high negative values during inspir-

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atory efforts against the obstructed upper airway <sup>9</sup>. Fletcher et al. have therefore pointed out that this «pharyngeal vacuum» and the consequent sucking back of the tongue play an important role in the pathogenic mechanism of complete glossoptotic pharyngeal obstruction <sup>9</sup>. They have recommended the insertion of a naso-pharyngeal tube to release the pharyngeal vacuum and the use of a long «lamb's nipple» to by pass the obstructing tongue for infants with congenital micrognathia <sup>9</sup>. Naso-pharyngeal and oro-pharyngeal tubes used to minimize the build-up of pharyngeal negative pressure have since been successfully used as a method to prevent glossoptosis.

### Methods which may prevent glossoptosis-apnoea in infants with urti

The genioglossus dysfunction. In some infants the genioglossus activity is impaired to such an extent that it is not able to counterbalance even the small increase of the aspirating force brought about by inspiratory efforts during URTI <sup>10</sup>. In 1923 this condition was first described by New in a 3-month-old infant with URTI and called «flaccid tongue» <sup>11</sup>. The associated dyspnoea be-

came worse during sleeping but lessened when a catheter was passed through the mouth into the pharynx. A celluloid appliance was therefore constructed with the same curvature and diameter of the catheter. When the celluloid piece was passed into the pharynx, the infant breathed around the tube without difficulty, even during sleep 11. Similarly, the use of a rubber oro-pharyngeal tube in infants who develope severe respiratory distress during URTI results in immediate relief of dyspnoea as this device depresses the tongue, thus «allowing ventilation through and around the tube» 12. Furthermore, clinical observations indicate that breathing with an open mouth is a protective manoeuvre against glossoptosis, whereas closing the mouth causes a glossoptotic pharyngeal obstruction in infants with genioglossus disfunction and URTI 13.

## Methods which may prevent glossoptosis-apnoea in infants with choanal atresia

The inability to mouth-breathing. There is still an held conviction that bilateral choanal atresia causes neonatal asphyxia because the infant is an obligatory nasal breather. In 1914 Richardson described a type of dispnoea charac-

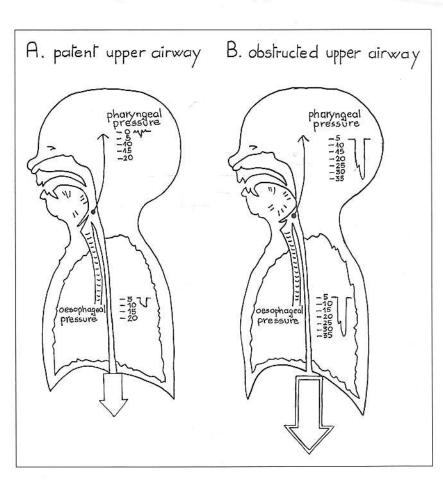


Fig. 1.
Diagram showing pressure tracings from hypopharynx and mid oesophagus during ventilatory phases without and with glossoptotic pharyngeal obstruction. Note increased negative pressure in pharynx associated with glossoptosis. Diagramma che mostra i tracciati delle pressioni rilevate a livello dell'ipofaringe e dell'esofago durante fasi ventilatorie senza e con ostruzione del faringe da glossoptosi. Notare le forti pressioni negative nell'ipofaringe associate a glos-

terized by alter bilateral choana of the infant to apnoea. The en he considered a thus relieving th depressing the between the lips ing and relieve «method of kee a simple nipple which the infan breathe without bility to open th efforts may caus A feeding tube the stomach mo billateral choun glosso-palatul « In 1977, Cozzii mouth-breathin of the pharynx infant with symp to oral ventilation airway resistan efforts 1-3 10 17 18 depression is tra results in a force backwards (Fig. by an appropriat the tongue will b it seals off the or ing the tongue w be a useful techn geal vacuum», tl apnoea in infants thermore, thumb a protective action sleep, some infa able to breathe without awakeni the obstructive with choanal atre effective in preve imize the «negat

## Possible mechan

In 1973, Swift a infants who were nasal oclusion by the dummies, but infants showed si these clinical obs gest that dummy to

terized by alternate apnoea and crying in an infant with bilateral choanal atresia 14. He believed that the inability of the infant to open his or her mouth was the cause of apnoea. The ensuing cyanosis stimulated crying, which he considered as a method to induce mouth-breathing, thus relieving the obstructive apnoea. The manoeuvre of «depressing the lower jaw and maintaining an opening between the lips» established satisfactory mouth-breathing and relieved the cyclic dyspnoea 14. A different «method of keeping the baby's mouth open» consists of a simple nipple with a large hole on its apex through which the infant with bilateral choanal atresia is able to breathe without respiratory distress (Fig. 2) 15. The inability to open the mouth and the consequent inspiratory efforts may cause the tongue to be sealed to the palate 16. A feeding tube which is passed through the mouth into the stomach may reduce the dyspnoea in infants with bilateral choanal atresia by breaking this obstructive glosso-palatal «seal» 16.

In 1977, Cozzi suggested that the infant's inability to mouth-breathing was mainly caused by an obstruction of the pharynx due to glossoptosis 1. In this view, the infant with symptomatic choanal atresia is able to switch to oral ventilation, but he or she experiences increased airway resistance which requires greater inspiratory efforts 1-3 10 17 18. The ensuing increase of the intrathoracic depression is transmitted to the pharyngeal cavity and results in a force which tends to aspirate the tongue backwards (Fig. 1). As this force is not counterbalanced by an appropriate increase of the genioglossus activity, the tongue will be progressively sucked backwards until it seals off the oro-pharyngeal airway 1-3 10 17 18. Depressing the tongue with a finger or the handle of a spoon can be a useful technique to break or to prevent «the pharyngeal vacuum», thus reversing or avoiding glossoptosisapnoea in infants with choanal atresia/stenosis 1210. Furthermore, thumb sucking with open lips may also have a protective action against glossoptosis-apnoea. During sleep, some infants with bilateral choanal stenosis are able to breathe through the mouth around the thumb without awakening. As soon as the thumb is removed, the obstructive apnoea recurs 10. Similarly in infants with choanal atresia, naso- and oro-pharyngeal tubes are effective in preventing glossoptosis-apnoea as they minimize the «negative pharyngeal pressure» 2 10.

# Possible mechanisms whereby nonnutritive sucking may stabilize the upper airway

In 1973, Swift and Emery noticed that five normal infants who were sucking dummies responded to digital nasal oclusion by breathing through the mouth around the dummies, but when the dummies were removed the infants showed signs of obstructive apnoea <sup>4</sup>. In 1979, these clinical observations induced Cozzi et al. to suggest that dummy use may protect from SIDS by keeping

the oral airway open, thus preventing a pharyngeal vacuum and the consequent sealing off the airway <sup>2</sup>. In 1984, Paludetto et al found that nonnutritive sucking was associated with an increase of transcutaneous oxygen tension in premature infants; they speculated that nonnutritive sucking may activate upper airway muscles and enhance alveolar ventilation <sup>19</sup>. In 1993, Mitchell et al. doubted that a dummy serves as an oral airway but suggested that dummy sucking may stimulate some sensorial receptors which increase the genioglossus muscle tone and help to maintain upper airway patency <sup>5</sup>. We are not aware of other studies concerning the possible mechanisms whereby the use of dummies may prevent episodes of partial or complete functional obstruction of the upper airway.

Clinical observations indicate that the methods that may prevent glossoptosis-apnoea are quite similar in infants with upper airway instability associated with different conditions including congenital micrognathia, hypertrophied adenoids, choanal atresia/stenosis, and URTI. These methods include opening the mouth, depressing

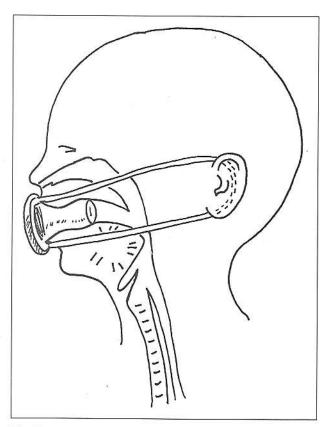


Fig. 2.

Illustration of the Mc Govern's nipple. This simple device (invented by the parents of a child born with bilateral choanal atresia) serves as an oral airway to relieve glossoptosis.

Illustrazione della tettarella di Mc Govern. Questa invenzione dei genitori di un bambino nato con atresia bilaterale delle coane è utile come cannula orale per evitare la glossoptosi.

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the tongue, thumb-sucking and the use of naso- or oropharyngeal tubes. The main physiological mechanism of these methods appears to be the establishment of an adequate oral or nasal airway, which avoids the pharyngeal vacuum and prevents vacuumglossoptosis apnoca. These clinical data, therefore, suggest that a dummy, like a thumb or a solid oro-pharyngeal tube, may serve as an oral airway which minimizes the build-up of negative pharyngeal pressure, thus reducing the aspirating force of the tongue.

Furthermore opening the mouth, depressing the tongue, thumb-sucking and the use of oro-pharyngeal tubes induce an opening of the mandible. According to Pierre

Robin, opening of the mandible is a manoeuvre that serves to displace the tongue forwards <sup>67</sup>. This manoeuvre may not appear protective because it implies a posterior displacement of the mandible angle, endangering the pharyngeal airway. Genioglossus muscle activity, however, increases when the jaw is either voluntary or involuntary opened <sup>20</sup>. Opening the mouth, therefore, stimulates a protective reflex to ensure a patent pharyngeal airway (Fig. 3). Opening the jaw more widely is associated with further enhancement of genioglossus activity <sup>20</sup>. These latter clinical and physiological data suggest that dummy sucking may protect from glossoptosis also by causing the mandible to open and thus

Enhancement of genioglossus activity dilates pharyngeal airway

1

Opening of the mandible stimulates receptors

2

Genioglossus inspiratory neuron excited

Fig. 3.
Schematic illustration showing one of the ways in which nonnutritive sucking may relieve glossoptosis.
Rappresentazione schematica di una delle vie attraverso cui la suzione non

nutritiva può evitare la glossoptosi.

Open-mouth breathing and/or thumb sucking
Observe during sleep, excitement, and respiratory tract infection
Symptoms and/or signs of glossoptosis-apnoea syndrome

NO
YES
Polysomnography
Obstructive apnoeas and desaturations
NO
YES

Associated apparent
life-threatening episode

NO
YES

Advise prone sleeping position
Glossopexy

Fig. 4. Algorithm for evaluation and manage ment of infants and children who are thumb suckers and/or open-mouth bre athers.

Algoritmo di procedure nei bambin che respirano con la bocca aperta e/o s succhiano il pollice.

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Open-mouth brhelp to identify airway instability glossoptosis apsleeping, or excinfants is imported SIDS 10. As open may reduce but believe that infa apparent life-that ic glossopexy 10.

<sup>1</sup>Cozzi F. Glossoptosis as cau (letter). Lancet 1977;2:830. <sup>2</sup>Cozzi F, Albani R

A common pathoph «The vacuum-gloss Med Hypotheses 19 <sup>3</sup>Cozzi F. Familial obstructive

N Eng J Med 1979; <sup>4</sup> Swift PGF, Emery *Clinical observation* Arch Dis Child 197

<sup>5</sup> Mitchell EA, Tayl Thompson JMD, S Roberts AP. Dummies and the su Arch Dis Child 1999. re that anoeuplies a langere activluntary erefore, oharynidely is glossus cal data lossop-

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stimulating the activity of genioglossus and other upper airway dilating muscles.

Tongue «median grooving» that forms a channel which serves as an oral airway and opening of the mouth are part of the particular manoeuvres of gasp, cough, and cry which the normal infant may adopt after birth to stabilize the upper airway and achieve respiratory autonomy 21. We conclude, therefore, that the above mentioned clinical observations support the concept that the protective action of dummy use against glossoptosis-apnoea may involve both the mechanisms previously postulated 2519. Dummy or thumb sucking with closed lips by opening the mandible stimulates a reflex enhancement of genioglossus which helps to maintain the patency of nasopharyngeal airway. Dummy or thumb sucking with open lips helps to establish an adequate oro-pharyngeal airway.

### **Implications**

Open-mouth breathing and thumb sucking may also help to identify those infants with a subclinical upper airway instability who present clinical manifestations of glossoptosis apnoeas/hypopnoeas only during URTI, sleeping, or excitement (Fig. 4). Identification of these infants is important because they are at increased risk of SIDS 10. As open-mouth breathing and thumb sucking may reduce but not eliminate the risk of SIDS, we believe that infants with an upper airway instability and apparent life-threatening episodes require a prophylactic glossopexy 10.

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