

*Review article*

Running title: Cleft Lip/Palate: Eating and Swallowing Disorders

## Eating and Swallowing Disorders in Children with Cleft Lip and/or Palate

Jelena Todorović<sup>1</sup>, Mirna Zelić<sup>2</sup>, Lana Jerkić<sup>3,4</sup>

<sup>1</sup>Health Center Zvečan, Zvečan, Serbia

<sup>2</sup>Center Terapika, Belgrade, Serbia

<sup>3</sup>Faculty of Special Education and Rehabilitation, Belgrade, Serbia

<sup>4</sup>Scholarship holder of Ministry of Education, Science and Technological Development of Republic of Serbia, Belgrade, Serbia

### SUMMARY

**Introduction.** Cleft lip and palate are complex congenital anomalies of the orofacial system of children. Feeding and swallowing problems occur with varying degrees in children with cleft lip and/or palate.

**Aim.** The aim of this paper was to review the literature and available evidence regarding the types of eating and swallowing disorders that can be identified in children with cleft lip and/or palate, as well as a description and types of compensatory strategies and interventions to alleviate difficulties.

**Methods.** Insight into the relevant literature was performed by specialized search engines on the internet and insight into the electronic database.

**Results.** The extent of the cleft is related to the severity of eating and swallowing disorders, so the most common problems are decreased oral sensitivity, cough, choking, nasal regurgitation, difficulty in sucking, laryngotracheal aspiration due to inadequate airway protection during swallowing, which may result in pneumonia and lung damage. Feeding and swallowing difficulty is also a source of stress for parents.

**Conclusion.** Choking, coughing, nasal regurgitation, laryngotracheal aspiration, excessive air intake can lead to dehydration, malnutrition, but also the need for alternative feeding methods. Therefore, it is of great importance to identify the problems of feeding and swallowing in a timely manner, along with modifications of the feeding method.

**Keywords:** cleft lip, cleft palate, eating disorder, dysphagia

Corresponding author:

Jelena Todorović

e-mail: jelena.milislavljevic90@gmail.com

## INTRODUCTION

Clefts of the orofacial region include a number of congenital anomalies, the most common of which are cleft lip and/or cleft palate. Most clefts occur under the influence of genetics, with the action of external factors and it develops in the early stage of pregnancy (1), between the fourth and ninth week of embryonic development (2).

Cleft lip and/or palate is characterized by discontinuity of the lips, hard and soft palate and alveolar bone (3). The severity of the clinical picture is determined by the lack of continuity of skin, muscle and bone tissue, and can manifest in a mild form such as damage of the lips, cleft lip and palate and alveolar bone, and in the most severe cases oblique facial cleft (3). The degree of difficulty and problems varies and depends on the degree and location of the split. Swallowing disorders can cause further nutritional or respiratory problems, creating significant stress for parents.

In children with cleft lip and/or palate, there are functional difficulties in swallowing, sucking, chewing, but also phonation, primarily due to changes in anatomical structures (4). The symptomatology of eating and swallowing disorders is diverse and, depending on the clinical picture, it can include: reduced oral sensitivity, cough, suffocation, nasal recruitment, difficulties in the sucking process, laryngotracheal aspiration.

## OROFACIAL CLEFT

On average, the incidence of orofacial cleft is 1:700, (5) but there are larger variations in incidence depending on geographical origin, race, socioeconomic status, but also environmental factors (6). In addition to genetics, several environmental factors are thought to contribute to it: intrauterine exposure to anticonvulsant therapy, folic acid deficiency, alcohol consumption, or smoking during pregnancy (2).

The spectrum of orofacial cleft is indeed wide (7). Due to the large variations of cleft lip, i.e. palate, from the morphological or clinical aspect, it is very difficult to establish a universal classification that is significant, both clinically and statistically (3). So far, several types of split classifications have been presented, however, the most common division is into four groups of splits. The first group involves cleft lip, unilateral or bilateral. The second group involves cleft lip and palate, unilateral or bilateral. The third

group includes cleft palate (palatoschisis), while the fourth group includes rare clefts, such as medial clefts of the lower lip, oblique cleft of the face, and transverse cleft of the face (8, 9). Ruptures of the orofacial region can also occur within the syndrome, and until today, over 400 syndromes are known that may include clefts (10).

The cleft lip and/or palate can be unilateral or bilateral. The cleft lip occurs due to the failure of the adhesion of the upper lip and the medial nasal part (11). A cleft palate may involve a cleft palate, a soft palate, or both, and a soft and hard palate when the uvula is often cleft. In some cases, the mucous membrane of the palate appears intact, but the muscles below are not properly formed, which is known as submucosal cleavage (11).

Velopharyngeal insufficiency occurs when there is an inability to close the sphincter between the oropharynx and nasopharynx, due to structural deficits (cleavage) or functional limitations (inadequate soft palate movements). The inability to close the velopharyngeal sphincter results in swallowing problems.

## SWALLOWING

Ingestion involves the act of forming a bolus in the oral cavity and its transit through the pharynx into the esophagus and stomach (12). This complex process requires precise coordination of more than 30 muscles located within the oral cavity, pharynx, larynx, and esophagus (13).

Different authors point to different divisions of swallowing phases. According to Logeman (14), swallowing has four phases: a) oral preparatory phase, b) oral, c) pharyngeal and e) esophageal. Matsuo and Palmer (15) additionally break down the oral phase into three more levels: food transfer through the oral cavity, food processing by chewing and saliva, and food transfer to the oropharynx. In the literature, one can often find a rough division into only three phases: oral, pharyngeal and esophageal.

Oral preparatory phase involves chewing movements and the formation of a bolus in the oral cavity. The patterns of movement in the oral preparatory phase vary, depending on the viscosity of the food, its quantity as well as the degree of pleasantness (subjective sense of taste) (16). The oral phase has the role of preparing food completely and facilitating the pharyngeal phase. The four inner

muscles of the tongue control the shape of the tongue (17), and the outer muscles control the position of the tongue (18). The tip of the tongue is raised, it touches the alveolar ridge, and the posterior part is lowered and opens a passage towards the pharynx. The dorsal surface of the tongue moves upwards, expanding the area of contact with the palate, pressing the fluid along the palate (19). The pharyngeal phase begins by inducing a pharyngeal swallowing reflex. The velopharyngeal sphincter rises and closes the path to the nasopharynx, the epiglottis closes and thus prevents food from penetrating inside the larynx and further into the airways (19). These actions achieve the separation of the digestive and respiratory tracts. The esophageal phase is the involuntary phase of swallowing.

The act of swallowing differs between newborns and adults. In newborns, the teeth are still not present, the hard palate is straight, and the hyoid bone and larynx are in a higher position than in adults (20). In newborns and young infants, none of the four phases of swallowing is voluntarily controlled, and only at a later age it will become the oral phase (21). In older children, chewing is a voluntary activity, relying on appropriate sensory bolus registration and motor response, and it is influenced by cognitive thought processes (22).

The oral phase in newborns is the primitive sucking reflex (23). The sucking reflex occurs when tactile stimulation occurs at the tip of the tongue or in the middle of the hard palate and on that occasion the newborn will move the tongue back and forth in a horizontal plane. Sucking reflexively triggers swallowing. For the first three months, infants fail to distinguish between liquid and solid and use the same sucking action for both (24). The sucking reflex appears at the beginning of the third trimester and lasts from 3 to 6 months after birth (25) when it is integrated into a more mature, voluntary sucking pattern. As the newborn develops, the tongue, lips, and lower jaw are able to achieve independent functions of biting, chewing, moving food, and forming bolus.

Since they use similar anatomical pathways, synchronization of breathing and swallowing is necessary to protect the airways and prevent aspiration. Suck-swallow-breath coordination enables efficient suction and swallowing with minimal impact on air flow (23). The consequences of aspiration can range from minimal to short-term, and to aspiration pneumonia or airway obstruction.

## SWALLOWING DISORDERS AND CLEFT LIP AND/OR PALATE

Swallowing disorders (dysphagia) include difficulty swallowing and controlling saliva, as well as feeding difficulties. In the broadest sense, dysphagia encompasses all behavioral, sensory, and preliminary motor actions in preparation for swallowing, including the awareness of the impending feeding situation, visual recognition of food, and increased saliva production as a physiological response to food (26).

Studies have shown the presence of eating and swallowing disorders in children with cleft lip and/or palate, but experts are not consistent in terms of symptoms (27). The most common symptoms of dysphagia are decreased oral sensitivity, delayed onset of swallowing, laryngotracheal aspiration causing coughing, choking, and vomiting during or after meals (28). Also, according to mothers, the most common problem is nasal reflux, suffocation and difficulties in the sucking process (29, 30). De Vries (31) observed that 86% of mothers of children with cleft palate did not even try to breastfeed. Choking, coughing, and excessive air intake can lead to dehydration, malnutrition, but also to the need for alternative feeding methods (31). Trenouth and Campbell (32) found that only infants with isolated cleft lip could maintain adequate weight only by breastfeeding. McDonald found in his study that the type of orofacial cleft had a significant effect on breastfeeding, as 60% of infants with cleft lip were breastfed for more than 6 months, while only a few newborns with cleft palate were breastfed for longer than 6 months (33).

With clefts of the orofacial region, there are difficulties in establishing efficient and safe sucking, due to weak intraoral pressure during sucking, but also difficulties in establishing suck-swallow-breath coordination (28).

Reid and co-workers (34) investigated the incidence of feeding problems in children with orofacial clefts and found that problems decreased with age, and the incidence of feeding problems decreased from 14 months to 15%. A study (27) showed that most feeding problems occurred when trying to breastfeed or bottle-feeding during the first months of life, and only in a small number of cases the problems did continue later with the introduction of solid and formed food.

Drowsiness during feeding could be attributed to higher energy consumption of the child in case of unsuccessful sucking, which leads to fatigue during feeding. Subsequent problems with biting and chewing could be explained, at least in part, by altered sensibility in the oral cavity due to orofacial cleft (27).

### **Cleft lip**

Newborns with an isolated cleft lip can breast-feed or use a standard bottle in a completely satisfactory way. The ability to suck remains adequate due to the intact soft palate. Although cleft lip may have a mild effect on labial adhesion around the nipple, breast tissue or an artificial nipple usually closes the cleft area (35). Children with isolated cleft lip showed a similar sucking pattern as their peers, while this pattern was completely altered in children with cleft palate with or without cleft lip (36). If the cleft lip also affects the alveolar ridge with a slight progression towards the front of the hard palate, it can affect the feeding process (35). The uneven surface of the alveolar ridge reduces lip adhesion and can lead to nipple movement within the oral cavity (35). The use of standard bottles or changes in position and cheek support may be helpful (37).

### **Cleft palate**

Ruptures involving the soft palate affect the creation of negative pressure during sucking due to the inability to close the velopharyngeal sphincter (35). One group of children has the ability to create negative pressure at the beginning of feeding, but cannot maintain it for the entire duration of feeding (36) and require the use of modified feeding bottles.

Due to the communication between the oral and nasal cavities, children with cleft palate may have nasal regurgitation and increased swallowing of air during feeding (35). Proper positioning and use of modified bottles will alleviate the problem.

In submucosal cleft palate, although the mucosa is intact, it is possible to expect the same problems as in open cleft. Therefore, they will also need to use modified bottles and proper positioning (37).

### **Cleft lip and palate**

The cleft lip and palate, whether unilateral or bilateral, eliminates the ability to create negative

pressure and reduces the force of compression (36). Extreme anatomical changes of the palate affect the movements of the tongue, which interferes with the capture of the nipple, with the possibility of nasal recruitment, since there is no closure of the velopharyngeal sphincter. A study (38) showed that the oral phase of swallowing is shorter, and the pharyngeal phase is longer in children with cleft lip and palate.

## **TREATMENT OF EATING AND SWALLOWING DISORDERS IN CHILDREN WITH CLEFT LIP AND/OR PALATE**

Services and treatment protocols for children with cleft lip and/or palate can vary greatly within and between developed countries. It is important to recognize that the treatment of children with orofacial clefts is long-lasting and complex, and since the birth of the child, various specialists have been involved who meet regularly and plan treatment together. Team members identify the resulting disorders, treat the child with orofacial cleft, or refer them to other specialists with more narrow knowledge (39).

Rehabilitation of these children is a complex and long-lasting process. Complete rehabilitation is possible only if it is started on time and if it is performed by a team of experts: orthodontist, pediatrician, surgeon, otorhinolaryngologist, speech therapist, psychologist (3). According to the WHO, the priorities are: surgical repair of various subtypes of orofacial clefts, surgical methods for the correction of velopharyngeal insufficiency, methods for the treatment of perioperative pain, swelling and infection (40).

One of the primary roles in the team is played by the orthodontist who determines the type, severity and morphological characteristics of the cleft (41). On the other hand, the speech therapist evaluates oral-motor abilities, readiness for feeding and safety of swallowing in newborns and children, and identifies the most appropriate feeding strategies and equipment to support long-term feeding development. Where appropriate, they perform an instrumental assessment of swallowing to perform an objective assessment of the swallowing function (42).

Techniques to facilitate feeding and swallowing in children with cleft lip and/or palate include positioning, oral relief techniques, assisted fluid delivery, the rate of fluid delivery, and changes

in fluid viscosity (43). Optimal positioning is the key to successful feeding, as it facilitates coordinated movements of the jaw, cheeks, lips and tongue during sucking and swallowing. Positioning a child with a cleft palate in an upright position of at least 60 degrees will allow gravity to help transfer fluid and swallowing. It also helps prevent nasal regurgitation, which can occur secondary to cleft palate (44). Stabilization of the jaw by placing the middle finger under the chin and forefinger between the chin and the lower lip helps to provide a stable platform for movements of the tongue, lips and cheeks (44).

Increasing the viscosity of the fluid to help maintain airway protection during ingestion is another often described intervention strategy with little objective evidence to support it. It is based on the change in the viscosity of the liquid, i.e. the use of condensed liquid, because the slower flow of liquid helps the child to protect the respiratory tract during swallowing (43).

There is a wide range of specialized nipples, bottles and cups available for use in newborns who have cleft lip and/or palate. Likewise, there is a wide range of independent descriptive studies related to these specialized subjects that report varying degrees of efficacy (43). Thus, artificial nipples are used, of different shape, length, size and thickness, but also different sizes of openings. The palatal obturator consists of an acrylic plate inserted into the mouth over the hard palate, basically closing the palate defect. Thus, separation of the nasal cavity and the oral cavity can be obtained (43). For children who are unable to suckle on their own, specialized bottles are used, which are used to deliver milk (45). The use of obturators is of great importance in feeding children with isolated cleft palate. By separating

the lip from the nasal cavity, it acts as a nipple resist during breastfeeding, reduces potential painful ulcerations of the nasal septum during swallowing, corrects adaptive positioning of the tongue in the cleft, expands it even more, improves airflow, restores the baby's ability to create intraoral pressure, retention of food in the cleft as well as its adhesion to the edges of the gape, and reduces nasal regurgitation and food congestion (46). In some cases, an oronasal fistula may remain after cleft palate surgery. If the oronasal fistula cannot be surgically closed, obturators are a permanent solution (47).

## CONCLUSION

Feeding and swallowing difficulty varies due to the severity in children with cleft lip and/or palate. Choking, coughing, nasal regurgitation, laryngo-tracheal aspiration, excessive air intake can lead to dehydration, malnutrition, but also to the need for alternative feeding methods.

Due to the complexity and long duration of rehabilitation, it is important to emphasize the importance of early treatment by a multidisciplinary team, which could prevent many problems that arise due to inefficient feeding. Early counseling, surgical intervention and selection of good feeding techniques is of great importance. Techniques to facilitate feeding and swallowing include positioning, oral relief techniques, assisted fluid delivery, fluid delivery rate, and fluid viscosity changes, using specialized nipples, bottles, and cups. Certainly, research should focus on developing a solid evidence base to support the effectiveness of the specific feeding interventions required for best clinical practice.

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# Poremećaji hranjenja i gutanja kod dece sa rascepima usne i/ili nepca

Jelena Todorović<sup>1</sup>, Mirna Zelić<sup>2</sup>, Lana Jerkić<sup>3,4</sup>

<sup>1</sup>Dom zdravlja Zvečan, Zvečan, Srbija

<sup>2</sup>Centar "Terapika", Beograd, Srbija

<sup>3</sup>Univerzitet u Beogradu, Fakultet za specijalnu edukaciju i rehabilitaciju, Beograd, Srbija

<sup>4</sup>Stipendista Ministarstva prosvete, nauke i tehnološkog razvoja Republike Srbije, Beograd, Srbija

## SAŽETAK

**Uvod.** Rascepi usana i nepca su složene, urođene anomalije orofacijalnog sistema dece. Problemi sa hranjenjem i gutanjem javljaju se u različitom stepenu kod dece sa rascepom usne i/ili nepca. Cilj. Cilj ovog rada je pregled literature i dostupnih dokaza u vezi sa vrstama poremećaja hranjenja i gutanja, koji se mogu identifikovati kod dece sa rascepom usne i/ili nepca, kao i opis i pregled kompezatornih strategija i intervencija za ublažavanje poteškoća.

**Metode.** Uvid u relevantnu literaturu izvršen je specijalizovanim pretraživačima na internetu i pristupom elektronskoj bazi podataka. **Rezultati.** Obim rascepa povezan je sa težinom smetnji u hranjenju i gutanju, pa su tako najčešći problemi: smanjena oralna osetljivost, kašalj, gušenje, nazalna regurgitacija, teškoće u procesu sisanja i laringotrahealna aspiracija, usled neadekvatne zaštite disajnih puteva tokom gutanja, što će za posledicu imati čak i upalu pluća i oštećenja pluća. Teškoće tokom hranjenja i gutanja su i izvor stresa za roditelje.

**Zaključak.** Gušenje, kašalj, nazalna regurgitacija, laringo-trahealna aspiracija i prekomerni unos vazduha, mogu dovesti do dehidracije, neuhranjenosti, ali i do potrebe za alternativnim načinima ishrane. Zato je od velikog značaja pravovremeno identifikovanje problema hranjenja i gutanja, uz modifikacije načina hranjenja.

**Ključne reči:** rascep usne, rascep nepca, poremećaj hranjenja, disfagija