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EATING AND SWALLOWING DISORDERS IN CHILDREN WITH CEREBRAL PALSY

POREMEĆAJI HRANJENJA I GUTANJA KOD DECE SA CEREBRALNOM PARALIZOM

Mile VUKOVIĆ[1] and Jelena TODOROVIĆ[2]

Summary

Introduction. Cerebral palsy is characterized by abnormalities of muscle tone, movement and motor abilities caused by disorders in brain development. Many children with cerebral palsy have severe swallowing problems. **Cerebral Palsy.** Cerebral palsy is a neurodevelopmental disorder characterized by abnormalities in brain development. Swallowing is a process by which a bolus that is formed within the oral cavity is transported through the pharynx into the esophagus and stomach. It has four phases: preparatory oral, oral, pharyngeal and esophageal. **Swallowing disorders in children with cerebral palsy.** Swallowing disorders vary depending on the type of cerebral palsy. Children with spastic paralysis have a more pronounced disorder of the voluntary phases of swallowing, while children with athetoid paralysis are expected to have a disorder at the oral-motor level. Neurological lesions may affect the muscle function of the jaw, cheeks, lips, tongue, palate and pharynx, which is manifested by difficulties in controlling saliva and swallowing. **Diagnosis and treatment of dysphagia in cerebral palsy.** Diagnosis and treatment of dysphagia is best accomplished by a team of experts including a pediatrician, neurologist, otorhinolaryngologist, gastroenterologist, radiologist, dental specialist, speech therapist, nutritionist, and a specialist in physical medicine and rehabilitation. **Conclusion.** Dysphagia is a common comorbidity in children with cerebral palsy. Although swallowing disorders often correlate with the type of cerebral palsy, the most prevalent signs of dysphagia include: coughing, prolonged feeding time, laryngotracheal penetration and aspiration, suffocation, which leads to dehydration, malnutrition, but also to the need for alternative feeding methods. It is important to emphasize the importance of early treatment by a team of experts, with a speech therapist as a team leader.

Key words: Cerebral Palsy; Child; Feeding and Eating Disorders; Deglutition Disorders; Diagnosis; Treatment Outcome

Sažetak

Uvod. Cerebralnu paralizu karakterišu abnormalnosti mišićnog tonusa, pokreta i motoričkih sposobnosti usled poremećaja u razvoju mozga. Mnoga deca sa cerebralnom paralizom imaju izražene smetnje u gutanju. **Cerebralna paraliza** je neurorazvojni poremećaj za koji su karakteristične abnormalnosti u razvoju mozga. **Gutanje** podrazumeva akt formiranja bolusa u usnoj duplji i njegov tranzit kroz ždrelo u jednjak i želudac. Ima četiri faze: pripremnu oralnu, oralnu, faringealnu i ezofagealnu. **Poremećaji gutanja kod dece sa cerebralnom paralizom** variraju u zavisnosti od tipa cerebralne paralize. Deca sa spastičnom paralizom imaju izraženiji poremećaj voljnih faza gutanja, dok se kod dece sa atetoidnom paralizom očekuje poremećaj na oralno-motoričkom nivou. Prisustvo neurološke lezije može da utiče na mišiće vilice, obraze, usne, jezik, nepce i ždrelo, što se manifestuje teškoćama u kontroli pljuvačke i u gutanju. **Dijagnostika i tretman disfagije kod cerebralne paralize** sprovodi se timski. Stručni tim uključuje pedijatra, neurologa, otorinolaringologa, gastroenterologa, radiologa, specijalistu dentalne medicine, logopeda, nutricionistu i specijalistu fizikalne medicine i rehabilitacije. **Zaključak.** Disfagija predstavlja značajan komorbiditet kod dece sa cerebralnom paralizom. Iako smetnje u gutanju često korelišu sa tipom cerebralne paralize, u opšte znake disfagije mogu se izdvojiti: kašljanje, produženo vreme hranjenja, laringotrahealna penetracija i aspiracija, gušenje, što dovodi do dehidracije, neuhranjenosti, ali i do potrebe za alternativnim metodama hranjenja. Važno je naglasiti značaj ranog lečenja u koje je uključen tim stručnjaka, pri čemu je logoped obično rukovodilac tima.

Ključne reči: cerebralna paraliza; dete; poremećaji hranjenja; poremećaji gutanja; dijagnoza; ishod lečenja

Introduction

Cerebral palsy (CP) is a neurodevelopmental disorder characterized by abnormalities in muscle tone, movement, and motor ability caused by disorders in brain development [1]. It is a heterogeneous group of nonprogressive motor disorders associated with muscle weakness, limited range of motion, spasticity, and pathological reflexes [2]. It is a consequence of various etiological factors that caused brain damage before or during birth, or in the first years of life [3].

In addition to impairments in motor skills, disorders of sensation, perception, cognition, speech and behavior, epilepsy and musculoskeletal disorders are also often observed in CP. Disorders of swallowing may also be present, leading to changes in the anticipatory, preparatory, oral, pharyngeal, and esophageal phases of swallowing [4]. Thus, eating and swallowing difficulties affect the nutritional status of persons with CP [5].

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Abbreviations
CP – cerebral palsy
HRM – high-resolution manometry

Cerebral Palsy

According to the current definition, developed by an international team of experts, CP is a group of permanent, but not unchanging, disorders of movement and/or posture and motor function, which are the result of non-progressive disorders, lesions or abnormalities of the developing brain [6]. The CP is one of the leading causes of neurological impairment in childhood [7]. The incidence of CP is 2 – 3 per 1000 term, live births, while the incidence in preterm infants it ranges from 40 to 200 per 1000 live births [3, 8, 9].

The etiology of CP is multifactorial, but in 92% of cases it is thought to occur in the perinatal period [10]. The risk factors include congenital malformations in the development of the cortex, hypoxia, asphyxia, while injury and central nervous system infections are the most common postnatal causes of CP [11]. Despite the identification of risk factors, in 80% of cases the cause of CP is unclear and it is considered idiopathic [12].

Due to the heterogeneity of the clinical presentations, CP is classified according to the type of disorder and the topography. From the clinical aspect and based on the type of motor disorder, spastic, dyskinetic, and ataxic types of CP are distinguished, while the hypotonic type is not included in the classification because it progresses into one of the three types [1].

The spastic type of CP is the most common, occurring in as many as 85% of cases [13]. It is characterized by increased muscle tone, increased resistance to passive movements, abnormal posture and movements, and scissor gait [14]. Instead of fine and individual movements, the movements are massive, while the voluntary movements are slower and are performed with effort [1]. This type of CP is further broken into three subtypes: 1) spastic hemiplegia, 2) spastic diplegia, and 3) spastic quadriplegia [1].

The dyskinetic type of CP occurs in about 7% of cases [13]. Motor disorders in this type are much more severe than in other forms, and are manifested by difficulties in performing targeted movements, uncontrolled movements, while repetitive movements are often observed [14]. Certain forms of non-motor comorbidity are also observed in CP, such as intellectual development disorders, epilepsy, impaired vision, hearing, and dysphagia [15]. The dyskinetic type of CP is manifested as choreoathetoid and dystonic type of CP [1].

The ataxic type of CP is the rarest and occurs in about 4% of cases [13]. It is manifested by decreased muscle tone, tremor, and disturbances in the performance of goal-directed movements [14]. In addition, children with the ataxic CP often exhibit abnormal patterns of posture or movement and loss of orderly muscle coordination, which affects the strength, rhythm, and accuracy of the arm, leg, and torso movements [16].

Swallowing and phases of swallowing

Ingestion is the process of bolus formation in the oral cavity and its transport through the pharynx into the esophagus and stomach [17]. This complex process requires precise coordination of more than 30 muscles located within the oral cavity, pharynx, larynx, and esophagus [18]. Understanding the normal physiology and pathophysiology of swallowing is the basis for the detection and treatment of swallowing disorders.

Different authors present different phases of the swallowing process. According to Logemann [19], swallowing has four phases: a) preparatory oral, b) oral, c) pharyngeal and e) esophageal. Some authors additionally divide the oral phase into three more phases: food transfer through the oral cavity, food processing by chewing and saliva, and food transfer to the oropharynx.

The preparatory oral phase involves chewing movements and the formation of a bolus in the oral cavity. The patterns of movement in the preparatory oral phase vary depending on the viscosity of the food, its quantity, as well as the degree of pleasure [20]. The oral phase has the role of preparing food completely and facilitating the pharyngeal phase. The tip of the tongue is raised, it touches the alveolar ridge, and the posterior part is lowered, opening the passage to the pharynx. The dorsal surface of the tongue moves upwards, expanding the area of contact with the palate and pressing the fluid along the palate [18]. The pharyngeal phase begins by inducing a pharyngeal swallowing reflex. The velopharyngeal sphincter rises and closes the path to the epipharynx, the epiglottis closes and thus prevents the penetration of food inside the larynx and further into the airways [18]. These actions separate the digestive and respiratory tracts. When it comes to swallowing fluids, this phase begins during the oral phase [21]. The esophageal phase is the involuntary phase of swallowing.

Ingestion consists of a series of activities that require highly integrated sensorimotor coordination of multiple structures: cranial nerves, muscles, centers in the brainstem, as well as bilateral areas in the cortex [22]. Studies have shown that the left hemisphere plays a greater role in motor planning and the components of voluntary swallowing. In contrast, the right hemisphere is more involved in the automatic aspects of the swallowing process, during the pharyngeal phase. Although the role of individual hemispheres in the act of swallowing has not been fully elucidated, empirical data testify to the existence of bilateral control of the swallowing process [22].

From the point of view of development, it can be said that in newborns and younger infants, none of the four phases of swallowing is voluntary and that voluntary control of the oral phase is established at a later age. In older children, chewing is a voluntary activity, relying on appropriate sensory bolus registration and motor response [23].

Swallowing disorders in children with cerebral palsy

Dysphagia is often seen in children with CP. It is a swallowing disorder that may occur at any stage, oral preparatory, oral, pharyngeal, or esophageal phase [24]. The incidence of dysphagia in children with CP ranges from 19 to 99%. It often results in inadequate food intake, aspiration, and subsequent respiratory tract infections [25]. Some authors believe that all people with CP have some gastrointestinal disorders at some point of life [26]. The variability in reporting the prevalence of dysphagia in CP probably reflects differences in the level of motor impairment, intellectual disability, and the presence of other comorbid impairments.

Lucchi et al. [27] reported presence of dysphagia in most people with CP. According to some data, about 66% of children with moderate dysphagia are on a specific semi-liquid diet, while 95% of those with severe dysphagia use alternative feeding methods. Difficulties in chewing solid food are present in as many as 70% of children with CP, and 60% have difficulties in drinking from a glass [28].

Empirical data show that the severity of swallowing disorders varies depending on the type of cerebral palsy [29]. The difference in the severity of swallowing disorders may be related to the extent and location of the neurological damage. Spastic paralysis occurs due to bilateral injury of the upper motor neuron (pyramidal and extrapyramidal pathway) and these children have a more pronounced disorder of the voluntary phases of swallowing. The athetoid type of CP occurs as a consequence of lesions of the basal ganglia, and in these children, the swallowing disorder occurs at the oral-motor level [29].

Proper oral skills, as well as coordination of the movements of the muscles of the orofacial region, especially the tongue, are required for the normal development of sucking, swallowing and chewing [30]. It is known that a neurological lesion associated with CP can affect the muscles of the jaw, cheek, lips, tongue, palate, and pharynx, which is functionally manifested by difficulties in saliva control, swallowing, and speech [31]. Bilateral damage to the upper motor neuron usually causes a swallowing disorder, which manifests itself in problems in the formation of food bolus and a delay in the transfer of solid and liquid food from the oral cavity to the digestive tract [32].

Studies about the characterization of dysphagia in CP point out that swallowing disorder occurs both due to problems in voluntary oral movements and in the reflex pharyngeal phase of swallowing. Signs of dysphagia, such as delayed onset and segmented swallowing (voluntary movement), have been shown to occur due to damage to cortical neural networks. On the other hand, the signs of disorders of the pharyngeal component of swallowing, i.e., automatic components of deglutination, such as throat clearing, suggest subcortical brain injury and/or basal ganglion necrosis [33].

It has also been shown that children with CP due to poor lip control show difficulties in receiving bolus and sucking. In addition, poor lip closure leads to food loss as well as excessive salivation [34]. Uncontrolled salivation, i.e., spillage from the mouth to the lips, chin, neck and clothes, is present in 40% of people with CP, and in 15% of cases, it manifests itself in a severe form [35]. Studies have shown that children with uncontrolled salivation have greater difficulty in forming a bolus, inability to completely close the lips, and more residual contents in the oral cavity after swallowing [36]. It is believed that there is a significant correlation between the inability to close the lips and uncontrolled salivation [37].

The spastic type of CP is also associated with spasticity of the respiratory musculature, rigidity of the chest wall, impaired posture and abnormal time of muscle activation [38]. These factors may also contribute to insufficient intrathoracic and subglottic pressure. It is therefore not surprising that children with CP use compensatory strategies while eating. The oral transit time in children with CP is longer if they have a severe motor impairment [5].

Empirical data show that disorders at any stage of swallowing affect the safety and efficacy of oral intake required for adequate nutrition and hydration [25]. Swallowing disorders correlate with prolonged meals, poor growth and nutritional status, and potential respiratory consequences [28].

Disorders during the pharyngeal phase may include delayed or incomplete airway closure during swallowing, or pharyngeal aspiration of food or fluid, and food debris in the pharynx [31]. Aspiration may occur before swallowing (due to incoordination of the tongue, which allows the bolus to spill prematurely over the base of the tongue or a delayed swallowing reflex); during swallowing (associated with inefficient laryngeal closure); or after ingestion [21]. In children with CP who have swallowing difficulties, silent aspiration is common, when food or fluid enters below the right vocal folds, without clinical signs and symptoms [31]. The overall incidence of pulmonary aspiration in children with CP due to oral motor dysfunctions is not precisely known, but frequent hospital admissions due to presumed aspiration pneumonia are reported [33].

The incidence of gastroesophageal reflux is estimated in over 50% of children with CP, which can be explained by lesions in the neuronal-anatomical swallowing center located in the medulla oblongata, which leads to reflex dysfunction [33]. Reflux episodes have been shown to cause not only gastrointestinal symptoms, such as regurgitation or vomiting, but also respiratory problems, such as recurrent respiratory infections, persistent cough, life-threatening episodes of apnea, and respiratory failure [39].

Diagnosis and treatment of dysphagia in cerebral palsy

Clinical and instrumental assessment is used to diagnose dysphagia in children with CP [40]. Clinical assessment includes evaluation of medical records, a thorough review of medical history, and heteroanamnesis.

Clinical assessment is often supported by instrumental diagnostic procedures [41]. Diagnostic methods of dysphagia in children are videofluoroscopy, flexible endoscopy, and if no other diagnostic intervention is possible, manometric assessment of swallowing is used [41].

Videofluoroscopy is considered the gold standard in the diagnosis of dysphagia. This method allows real-time insight and visualization of the bolus through the oral cavity, oropharynx, hypopharynx, and esophagus, using modified barium [42]. It also provides information on the existence of absorption/penetration into the airways (before, during and after ingestion), the amount and localization of food residues in the oral cavity and pharynx [41].

The next diagnostic procedure is *flexible endoscopy*, which is used to assess the structures and functions of the upper respiratory tract, secretions, as well as pharyngeal phase of swallowing. It is performed using a flexible laryngoscope placed through the nose and then through the pharynx, in order to see the pharyngeal and laryngeal structures during swallowing [43]. It enables recording of the superior part of the pharynx and larynx. The obtained are color images [41].

One of the diagnostic methods is *high-resolution manometry (HRM)*. This method is used to objectively measure the swallowing pressure generated by muscle force along the pharynx and esophagus. The main indication for using HRM is the impossibility to assess swallowing differently, in cases of aspiration, nasal regurgitation, and coordination of breathing and swallowing [41].

After detecting signs of swallowing disorders and diagnosing dysphagia, children are included in the appropriate treatment program. Treatment of dysphagia in children with CP is performed by a professional team that includes doctors of appropriate specialties (pediatrician, neurologist, otorhinolaryngologist, gastroenterologist, radiologist, and dental specialist), speech therapist, nutritionist and a specialist in physical medicine and rehabilitation. Due to the nature of swallowing disorders, a speech therapist is usually the supervisor of the therapeutic process [41]. A speech therapist assesses oral-motor abilities and determines the most appropriate feeding techniques [40]. Also, the speech therapist, in cooperation with the physiotherapist, determines the best position when feeding the child with CP. From the therapeutic aspect of swallowing disorders in children, techniques for strengthening the muscles of the lips, tongue and jaws [44], as well as proper positioning during feeding, are most important. Also, impaired hand movement with inadequate body posture may disable self feeding, so they often need parental support or the support of a therapist [45]. Parental attitude towards the child's diagnosis is very important for the success of therapeutic interventions [46].

Conclusion

Data on the incidence of dysphagia in children with cerebral palsy vary widely and range from 19% to 99%. Despite the lack of precise data on the prevalence, it should be noted that most empirical studies show that dysphagia is a significant comorbid disorder in children with cerebral palsy. It has also been shown that eating and swallowing disorders vary in severity and depend on the type of cerebral palsy. Dysphagia in children with cerebral palsy is characterized by a whole set of different signs, some of which occur due to problems in the oral phase, and others in the pharyngeal phase of swallowing. Some signs of swallowing disorders, such as choking, coughing, prolonged feeding time, laryngotracheal penetration and aspiration, can lead to dehydration, malnutrition, but also the need for alternative feeding methods. Due to the complexity of the disorder, it is important to emphasize the importance of early diagnosis and treatment of swallowing disorders. Early detection of disturbances and early intervention in the area of feeding and swallowing can prevent many problems. Early counseling with parents and timely selection of good feeding techniques are of particular importance for the treatment of dysphagia in children with cerebral palsy. Although different techniques are used in the treatment of dysphagia in children with cerebral palsy, empirical data show that there is still no solid, scientifically verified evidence of the effectiveness of existing interventions in the field of eating and swallowing disorders in children with cerebral palsy. Therefore, future research should be directed towards obtaining as objective evidence as possible about the effects of existing therapeutic methods. Part of future research in this area should be dedicated to the development of new therapeutic procedures. We are sure that the implementation of appropriate therapeutic interventions leads to an improvement in the quality of life of children and their family members. Also, improvement of eating and swallowing contributes to better emotional and physical health of these children.

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