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Mini Review

Dysphagia in Cerebral Palsy

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ABSTRACT

Introduction: Cerebral palsy is the most common physical disability of childhood. Dysphagia is characterized by impairments in one or more of the following stages of swallowing: oral, pharyngeal and esophageal. The prevalence of dysphagia in children with cerebral palsy is 50.4%, with greater risk in those with more severely impaired functioning.

Objective: To conduct a scoping review on dysphagia in children with cerebral palsy regarding its prevalence, pathophysiology, specific signs and symptoms, red flags, screening, clinical evaluation, videofluoroscopic swallowing study features, functional classification, caregivers burden and treatment.

Data source: The search was carried out in May 2021 in the PubMed; Medline; and SciELO databases.

Data synthesis: Overall, 703 articles were found in the initial search process. Of these, 514 duplicates were removed. Then, 117 were excluded due to lack of appropriateness, thus yielding a total of 72 studies included in the scoping review.

Conclusion: Dysphagia can negatively impact many dimensions of the patient's health, and the quality of life of the patient and family. Dysphagia is the main risk factor for tracheal aspiration, and consequently for aspiration pneumonia and chronic lung disease, also influencing the risk of death because aspiration pneumonia is the main cause of death in all cerebral palsy age groups. Early recognition and adequate management of dysphagia are very important in order to identify the swallowing impairment, to improve swallowing skills and to prevent chronic lung disease and premature death. The interdisciplinary participation of medical specialties and rehabilitation professionals is essential for the assessment and treatment of other clinical, neurological and neurodevelopmental comorbidities that can negatively interfere with the swallowing skills and with the risk of tracheal aspiration. For the indication of any treatment, one should take into account, in addition to health issues, the socioeconomic and cultural characteristics of each family.

ABBREVIATIONS

CP: Cerebral Palsy; QoL: Quality of Life; LP: Laryngeal Penetration; OPA: Oropharyngeal Aspiration; CLD: Chronic Lung Disease; PRISMA-ScR: PRISMA Extension for Scoping Reviews; CNS: Central Nervous System; OA: Overt Aspiration; SA: Silent Aspiration; GMFCS: Gross Motor Function Classification System; EDACS: Eating and Drinking Ability Classification System; FEES: Fiberoptic Endoscopic Evaluation of Swallowing; VFSS: Videofluoroscopic Swallowing Study; GERD: Gastro-Esophageal Reflux Disease; VAS: Visual Analogue Scale; SOMA: Schedule for Oral

Motor Assessment; DDS: Dysphagia Disorders Survey; PSAS: Pre-Speech Assessment Scale; MBSS: Modified Barium Swallow Study; MBSImP: Modified Barium Swallow Impairment Profile

INTRODUCTION

Cerebral Palsy (CP) is the most common physical disability of childhood [1]. CP is defined as a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by epilepsy, secondary musculoskeletal problems, and disturbances of sensation, perception, cognition and communication [2,3]. Dysphagia is characterized by impairments in one or more stages of swallowing: oral, pharyngeal and esophageal [4]. The prevalence of dysphagia in CP is 50.4%, with greater risk in those with more severely impaired functioning [5]. Persons with CP experience many restrictions in eating and drinking, leading to lower self-esteem, poor quality of mealtime experiences, social isolation and depression [5,6]. The clinical and psychosocial consequences of dysphagia affect the child and the family, influencing their Quality of Life (QoL).

Laryngeal Penetration (LP) is the passage of food into the larynx but not through the vocal folds [7]. Oropharyngeal Aspiration (OPA) is the recurrent aspiration of saliva, food, and/or fluids below the level of the vocal folds [8]. Dysphagia is the main risk factor for OPA, and consequently for aspiration pneumonia, for Chronic Lung Disease (CLD), and can also influence the risk of death in children with CP. Most deaths in children with CP are attributed to diseases of the respiratory system (12 times higher in relation to the general population), with aspiration pneumonia as the main cause in all age groups [9-11]. Given its clinical and psychosocial relevance, this study aims to conduct a scoping review on dysphagia in CP.

MATERIALS AND METHODS

The study is characterized by a scoping review, based on the criteria of the PRISMA Extension for Scoping Reviews (PRISMA-ScR) [12].

Search strategy

The search for articles was carried out in May 2021 in the PubMed, Medline, SciELO, and LILACs databases, using the last 30 years as a filter and the keywords "cerebral palsy" and "dysphagia".

Recruitment and selection bias

The identification of articles for the study flowchart included four phases: (a) identification (study recruitment); (b) selection (duplicates); (c) eligibility (exclusion due to absence of eligibility according to the inclusion criteria) and (d) inclusion (inclusion of the remainders in the scoping review). Each phase was performed separately by two researchers who independently examined the title, the abstract and, when available, the main text of each study.

Inclusion criteria and study characteristics

The following inclusion criteria were used: to be directly related to the objectives of the review; to include children and adolescents of both genders, aged 0 to 18 years; to be published in English, Portuguese, Spanish and French; to be available in full text.

Results

Overall, 703 articles were found in the initial search process. Of these, 514 duplicates were removed. Then, 117 were excluded due to lack of appropriateness after reading the titles and abstracts and/or the full-text articles (for those with full text available), thus yielding a total of 72 studies included in the scoping review, as shown in the flowchart (Figure 1).

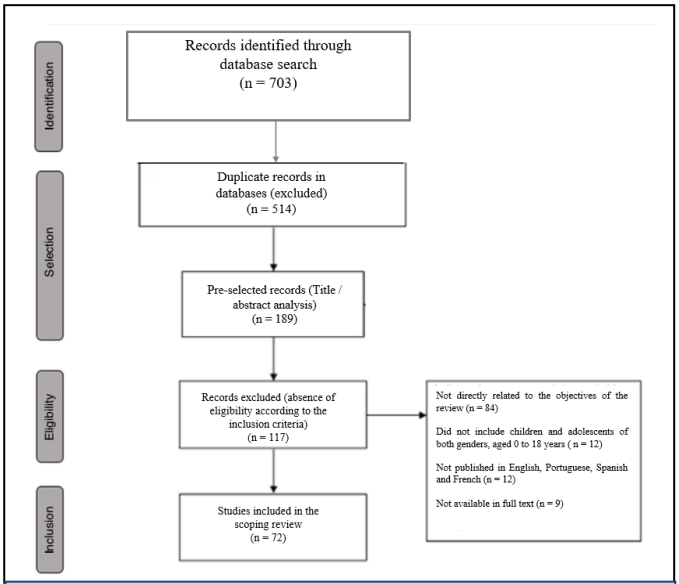

Figure 1: Study eligibility flowchart according to PRISMA-ScR.

DISCUSSION

Physiology

Normal gastrointestinal tract function results from a balanced interaction between the enteric nervous system and the Central Nervous System (CNS) which is called “the brain–gut axis” [13].

Swallowing is a highly complex process composed by a synergistic neuromuscular action involving food preparation, formation of the bolus, oral transit and propulsion of the bolus through the pharynx with protection of the airways, organized in sequentially related three stages: the oral, pharyngeal and esophageal phases [14].

Pathophysiology

In individuals with CP, the lesions often compromise the cortical and subcortical regions responsible for the harmonic functioning of the digestive system, leading to problems in both the volitional oral movements and the more reflexive pharyngeal phase of swallowing. Moreover, impaired ability to plan and coordinate swallowing with ventilation are consistent with brainstem involvement [13,14]. Overt Aspiration (OA) is the laryngeal cough reflex, whereby the aspirated material stimulates mechanoreceptors or chemoreceptors of the hypopharynx and laryngeal aditus and generates a reflexive cough in an attempt to expel the aspirated material. Silent Aspiration (SA) is the occurrence of aspiration in the absence of cough/other response. Most cases of aspiration involve SA [8].

The pathological vicious cycle of intervening comorbidities

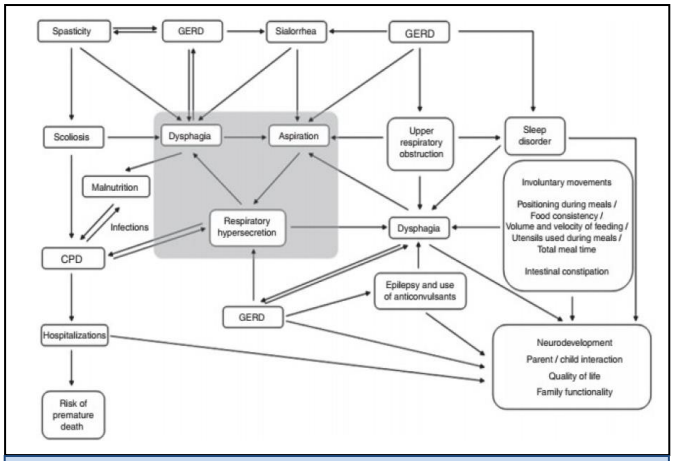

Figure 2: The pathological vicious cycle of intervening comorbidities [15].

The clinical and psychosocial consequences generated by dysphagia in CP suffer direct interference, both in frequency and in severity, due to the presence of other clinical, neurological and neurodevelopmental comorbidities. Conversely, the presence of dysphagia also interferes with the frequency and severity of these other comorbidities. In other words, the clinical, neurological and neurodevelopmental comorbidities in CP act intervening on each other, generating a pathological cycle of cumulative consequences called "the pathological vicious cycle of intervening comorbidities" [15] (Figure 2).

Dysphagia, aspiration and respiratory hypersecretion (due to aspiration) are the core of this vicious cycle. The objectives of the schematic figure are to show that: (1) spasticity, scoliosis, CLD, GERD, sialorrhea, upper respiratory obstruction, sleep disorder, epilepsy and use of anticonvulsants, involuntary movement, positioning, food consistency, volume and speed of food offering, utensils used in meals and intestinal constipation interfere in the dysphagia / aspiration / respiratory hypersecretion cycle; (2) these comorbidities can also negatively interfere (they are intervening) among themselves, exacerbating the dysphagia / aspiration / respiratory hypersecretion cycle; (3) the dysphagia / aspiration / respiratory hypersecretion cycle generates malnutrition, infections, CLD and hospitalizations, interfering with neurodevelopment, parent / child interaction, QoL and family functionality, and increasing the risk of premature death; (4) the approach to dysphagia is not limited to the difficulty of swallowing itself, but to all comorbidities and conditions that interfere in the dysphagia / aspiration / respiratory hypersecretion cycle.

Like all of us, patients with CP are also unique organisms in which multiple systems interact dynamically in search of a balance called homeostasis. Failures in one system, depending on the nature and extent of the failure, can affect that system in isolation or can cause imbalances in other systems, or in the organism as a whole. CP is a prototype in which brain damage can generate, or make it more likely, failures not only in the CNS, but also in several other systems, in a cascade of dynamically intervening comorbidities.

CP functional classification

CP is classified in how the deficiencies affect activity and participation [16]. There are classification systems for gross motor (Gross Motor Function Classification System - GMFCS), manual, communication, eating / drinking (Eating and Drinking Ability Classification System - EDACS) and visual skills and functions [17-21]. These classifications, emphasize the need to describe children's abilities rather than their disabilities, with a focus on how they habitually behave in real-world situations. GMFCS and EDACS are shown in (Table 1).

Table 1: GMFCS and EDACS[21].

Scale / Level	I	II	III	IV	V
GMFCS	Walks without limitations	Walks with limitations	Walks using a hand-held mobility device	Self-mobility with limitations; may use powered mobility	Transported in a manual wheelchair
EDACS	Eats and drinks safely and efficiently	Eats and drinks safely but with some limitations to efficiency	Eats and drinks with some limitations to safety; there may be limitations to efficiency	Eats and drinks with significant limitations to safety	Unable to eat and drink safely – tube feeding may be considered to provide nutrition

Dysphagia, GMFCS and EDACS

Kim JS et al (2013) [22] performed Videofluoroscopic Swallowing Studies (VFSS) in 29 children with CP. Nearly all of the children GMFCS III, IV and V had simultaneous abnormalities in the three stages of swallowing. Fifty percent of the children GMFCS IV and V demonstrated significant aspiration, but only 14.3% of the children GMFCS III and none of those with GMFCS I and II. García Ron A et al (2020) [23] assessed the swallowing skills of 44 children with CP. While the majority of patients below GMFCS III were EDACS I, 100% with GMFCS V were EDACS ≥ III. Benfer KA et al (2013) [24] studied 120 infants with CP. There were an increasing number of children with dysphagia for each increase in GMFCS level, these relationships were generally significant for children in GMFCS III to V compared with GMFCS I. All children who were tube fed were GMFCS IV and V. Speyer R et al (2019) [5] performed a systematic review and metanalyses on prevalence of drooling, swallowing, and feeding problems in CP. Pooled prevalence of dysphagia estimates stratified by GMFCS level were: 16.3% for level I, 51.7% for level II, 60.4% for level III, 84.2% for level IV, and 97.9% for level V. Pooled prevalence estimates for GMFCS levels I to III combined were 23.9% and 88.3% for GMFCS levels IV to V combined.

Dysphagia and Gastro-esophageal reflux disease (GERD)

GERD and dysphagia alone or in comorbidity are common in children with CP. GERD can be attributed to hiatus hernia, adoption of a prolonged supine position, and increased intraabdominal pressure secondary to spasticity, scoliosis or seizures [24,25].

Table 2: Main signs and symptoms of GERD and dysphagia in CP[25,27-29].

GERD		
Signs and symptoms	**Infants**	**Older children**
Common manifestations	- Regurgitation (especially postprandially) - Signs of esophagitis (irritability, arching, choking, gagging, feeding aversion) - Failure to thrive - Sandifer syndrome	- Regurgitation during the preschool years - Abdominal and chest pain in later childhood and adolescence - Sandifer syndrome
Respiratory manifestations	- Obstructive apnea or as stridor or lower airway disease in which reflux complicates primary airway disease such as laryngomalacia or bronchopulmonary dysplasia, Otitis media, sinusitis, lymphoid hyperplasia, hoarseness, vocal cord nodules, and laryngeal edema	- More commonly related to asthma or to otolaryngologic disease such as laryngitis or sinusitis
Dysphagia		
Signs and symptoms		
Impairments in swallowing phases		
Oral phase	- Insufficient suck - Incomplete lip closure - Drooling - Inefficient capturing of food bolus - Food falls from the mouth	- Tongue action ineffective in forming bolus - Pooling of bolus in lateral sulci - Prolonged bolus formation - Insufficient chewing - Inability to clear the oral cavity
Pharyngeal phase	- Difficult swallowing - Repetitive swallowing - Inadequate laryngeal elevation - Coughing /choking at meal times - Wet breathing - Gagging	
Esophageal phase	- Hyperextension of head - Food refusal - Regurgitation - Nasal reflux - Nighttime awakening	- Vomitus on pillow - Vomiting - Acidic–smelling breath - Unexplained irritability surrounding mealtimes
OPA (OA and SA)	- Increased congestion at meal times - Tachypnea / dyspnea at meal times - Tearing at meal times - Gurgly voice at meal times - Cianosis at meal times - Recurrent pulmonary infection	
Systemic	- Lack of weight gain over 2–3 months in young child, not just weight loss	
Others	- Prolonged meal time with insufficient consumption	

Nevertheless, CNS dysfunction is likely to be the prime cause of GERD. As a result of neuromuscular incoordination, the anti-reflux function of the lower esophageal sphincter mechanism

and esophageal motility are significantly impaired [25,26]. Asgarshirazi M et al (2017) [25] studied 50 children with CP, 73% of patients with GMFCS IV and V showed concurrence of GERD and dysphagia and only 21% in mild gross motor dysfunction. In agreement with other studies, patients had fewer pneumonia and choking after GERD treatment. Physicians should pay attention to GERD in CP because early treatment can prevent severe complications. The main signs and symptoms of GERD in CP are shown in (Table 2) [25,27].

Dysphagia specific signs and symptoms in CP

The signs and symptoms of dysphagia in CP can emerge from functional impairments in swallowing phases (oral, pharyngeal and esophageal), from OPA (OA and SA) or can be systemic (Table 2) [28]. Such signs and symptoms should be actively investigated through anamnesis, physical examination, direct clinical evaluation of swallowing and VFSS or Fiberoptic Endoscopic Evaluation of Swallowing (FEES).

Red flags

Red flags for dysphagia, undernutrition, and respiratory complications, increase the chances of reduced QoL, hospitalizations, indication for gastrostomy and risk of premature death. These red flags are shown in table 3 and must be actively investigated [4,7,30-33].

Table 3: Red flags for dysphagia, undernutrition, and respiratory complications [4,7,30-33].

Dysphagia	Undernutrition	Respiratory complications
- Average meal duration ≥30 minutes on ≥2 days in a 3-day food record	- Physical signs of undernutrition	- LA
- Moderate meal stress (child / caregiver) in a scale where 1-2=None-mild, 3-5=Moderate-severe	- Weight for age z score < 2	- OPA (OA and SA)
- Lack of weight gain over 2–3 months in young child, not just weight loss	- Triceps skinfold thickness < 10th centile for age and sex	- Abnormal esophageal body function
- Increased congestion at meal times, “gurgly” voice, respiratory illnesses	- Mid-upper arm fat or muscle area < 10th centile	- GERD
- GMFCS III-V	- Faltering weight and/or failure to thrive	– Increased PaCO2

Screening

Bell KL et al (2019) [34] validated a screening tool for feeding / swallowing difficulties and undernutrition in children with CP that can be used independently by parents / caregivers. The tool successfully identified 100% of children with severe undernutrition and 100% of children in EDACS level IV or V. The 4-item tool (Table 4) total score of ≥3 refers for further assessment of feeding / swallowing and nutritional status.

Table 4: Screening tool for feeding / swallowing difficulties and undernutrition in children with CP[34].

Question	Possible response and scoring
Do you think your child is underweight?	Yes (1), No (0), Unsure (1)
Does your child have problems gaining weight?	Yes (1), No (0), Unsure (1)
Rate, on a scale from 0–10, whether you think your child has any problems eating compared to other children of his / her age	10cm long VAS with numbers at each centimetre ≥7 on the VAS=score 1
Rate, on a scale from 0–10, whether you think your child has any problems drinking compared to other children of his / her age	10cm long VAS with numbers at each centimetre ≥7 on the VAS=score 1

Evaluation

Dysphagia should be suspected and assessed in CP in all children with criteria based on the screening tool for feeding / swallowing difficulties and undernutrition [34], those with GMFCS levels III, IV and V and / or those with red flags for dysphagia in order to improve swallowing skills, growth and nutritional outcomes and respiratory health [4,23,24]. The main objectives in the assessment of dysphagia in CP are clearly to answer the following questions: (1) What are the indicated motor learning interventions?; (2) Are there indications for compensatory strategies (positioning, food textures, utensils, volume and rhythm of feeding) to make oral feeding more functional, pleasurable and safe?; (3) Are the time and energy required of the caregiver and the child to finish a oral-feeding meal rewarding in terms of QoL?; (4) The current procedure by which the child is fed is sufficient to ensure satisfactory hydration and nutrition? (5) Are there intervening comorbidities acting to worsen the severity of dysphagia? (6) Is the oral-feeding safe and does not lead to aspiration risk?; (7) Is there an indication for enteral tube-feeding regimens?

Table 5: Roles of each professional directly involved in the dysphagia assessment[4,33,35-43].

	Attending physician		Speech therapist		Nutritionist	Nurse
Anamnesis	- Active investigation of comorbid symptoms: spasticity, intestinal constipation, GERD, sialorrhea, pulmonary hypersecretion, upper respiratory obstruction, sleep disorder, recurrent upper and lower airway infections, hospitalizations, epilepsy, medication use, neurodevelopment, pain		- Red flags - Persons involved with feeding; differences in feeding styles - The type, texture, viscosity, quantity, and quality of the food - The timing, frequency, and duration of meals - The feeding environment, distractions - The feeding routine, technique (volume and offering rhythm), adaptive equipment, positioning - GERD signs, anterior and posterior sialorrhea signs		- 3-day-weighed food record	- Evaluation of intestinal function - Assessment of the caregiver's difficulties in daily patient care - Social, economic and psychological family characteristics
Physical exam	Before the evaluation of the speech therapist	- Active investigation of signs of comorbidities: complete physical, neurological and orthopedic examination	Before the evaluation	- Positioning for feeding (patient and caregiver); utensils; mouth opening; lip seal; tongue mobility; oral and oropharyngeal sensitivity; voice; drooling; choking before swallowing	- Anthropometric measurement (weight, height, BMI, triceps skin fold, mid-arm circumference) - Classification of nutritional status using CP-specific growth charts stratified by gender, tube or oral feeding status and functional severity (GMFCS I to V)	- Clinical respiratory monitoring and by oximetry
			Throughout the evaluation	- Positioning for feeding (patient and caregiver); utensils; food textures offered; volume and rhythm during the offer - SOMA, DDS or PSAS		
	Throughout the evaluation of the speech therapist	- Clinical respiratory monitoring and by oximetry	Oral phase	For each food texture and liquids: - Mouth opening; lip seal; sucking; captation; fall out of mouth; formation of the bolus; chewing; ability to clean the oral cavity; drooling		
	After the evaluation of the speech therapist	- Respiratory status	Pharyngeal phase	For each food texture and liquids: - Delayed swallowing; multiple swallows; oral cavity food stasis, coughing/choking		
			Esophageal phase	Hyperextension of the head; nasal reflux		
Complementary exams	- According to the need in cases in which comorbidities were detected through anamnesis and physical, neurological and orthopedic examination		VFSS indications		- According to the need in cases of nutritional concern	- According to the need in cases in which comorbidities were detected through anamnesis and physical, neurological and orthopedic examination
			Scenarios where the exam is easily available	Virtually generalized indication		
			Scenarios where the exam is not so available	Risk of aspiration (by history or observation), prior aspiration pneumonia, suspicion of a pharyngeal or laryngeal problem (for example, breathy or husky voice quality), gurgly voice quality		
Conduct / treatment	- Initial and specific treatment for each comorbidity - Referral for joint or complementary assessment with other medical specialties (for interpretation of complementary exams and more complex specific procedures) or with other professionals in the rehabilitation team (more complex specific procedures) - VFSS indication - Indication of enteral tube-feeding regimens - Joint team discussion		- EDACS classification - Motor learning interventions - Compensatory strategies - VFSS indication - Indication of enteral tube-feeding regimens - Joint team discussion		- Recommendations regarding nutrition and hydration, and growth patterns - Indication of enteral tube-feeding regimens - Joint team discussion	- Counseling on intestinal reeducation - Counseling to facilitate the caregiver's difficulties in daily patient care - Joint team discussion

Comprehensive evaluation

A comprehensive assessment requires the active search for signs and symptoms of dysphagia itself, its clinical and psychosocial consequences and intervening comorbidities. The full assessment, ideally, should be carried out jointly by the attending physician, the speech therapist, the nutritionist and the nurse. Each one will evaluate through anamnesis (in a single and joint questionnaire), physical examination, direct clinical evaluation of swallowing and complementary exams in order to

answer all the questions mentioned above. Participation and opinions of the patient and family are essential in this process. Depending on the needs of each patient-family set (intervening comorbidities, child's physical characteristics and psychosocial needs of the family), case discussions and actions by other medical specialties (gastroentrologist, pulmonologist, otolaryngologist, allergist, neurologist, gastrosurgeon, orthopedist, neurosurgeon) and by other rehabilitation professionals (physiotherapist, occupational therapist, psychologist and social worker) may be necessary. Table 5 details the roles of each professional directly involved in the assessment [4,33,35-41].

Non-invasive objective clinical standardised dysphagia measures

The Schedule for Oral Motor Assessment (SOMA) for children aged between 8 and 24 months, the Dysphagia Disorders Survey: Pediatric (DDS) for children and adults with a developmental disability, and the Pre-Speech Assessment Scale (PSAS) for examining 27 pre-speech feeding behaviour are the most appropriate non-invasive objective clinical measures [35,36].

Videofluoroscopy Swallowing Study (VFSS), Modified Barium Swallow Study (MBSS) and Modified Barium Swallow Impairment Profile (MBSImP)

VFSS is a Modified Barium Swallow Study (MBSS) that permits real-time dynamic visualization of bolus flow and structural movement throughout the upper aerodigestive tract using videofluoroscopic imaging. Currently VFSS is considered the gold standard method for examining oropharyngeal swallowing physiology and impairment, and further allows for detection of the presence, timing, depth, and patient response to airway invasion (penetration / aspiration) while assisting in the identification of the physiologic cause of swallowing impairment [42-46].

The Modified Barium Swallow Impairment Profile (MBSImP) is a research-based, standardized, and validated approach for interpretation and quantitation of MBSS findings. The MBSImP provides quantitative evaluation of 17 physiologic components of swallowing distributed across three functional domains: oral, pharyngeal, and esophageal. MBSImP components are scored on an ordinal scale from 0 (indicating no impairment) to a maximum of 2, 3, or 4, depending on the specific component. The MBSImP scoring instrument is composed of six measures of oral impairment (lip closure, tongue control during bolus hold, bolus preparation / mastication, bolus transport / lingual motion, oral residue, initiation of pharyngeal swallow, 10 measures of pharyngeal impairment (soft palate elevation, laryngeal elevation, anterior hyoid excursion, epiglottic movement, laryngeal vestibular closure–height of swallow, pharyngeal stripping wave, pharyngeal contraction, pharyngoesophageal segment opening, tongue base retraction, pharyngeal residue), and one measure of esophageal impairment (clearance in the upright position) [44,45,47].

Caregivers

Dysphagia affects the caregivers' QoL. In some cases caregivers spend an excess of three hours per day feeding the CP patient, with individual mealtimes lasting more than half an hour [48]. Social support is considered to be a buffer against the stress experienced by caregivers [49]. The support needs of families and caregivers should be assessed, including the values parents attach to oral and tube feeding. Structured support should be offered with appropriate training in order to ensure parental information needs, and any emotional, practical and financial issues to be addressed [48].

Treatment

In practice, to indicate any treatment in children with CP, one should take into account the socioeconomic and cultural characteristics of each family, in order to be efficient, specific, and less of a burden for each patient / family [15]. As already described, the treatment of dysphagia is not limited to the therapeutic modalities directly related to dysphagia. Clinical and psychosocial comorbidities should be actively researched, diagnosed and treated appropriately. Hence the relevance of multidisciplinary participation.

The direct interventions for dysphagia can be grouped into two categories; those that align with motor learning principles, which aim to improve skills, and those that primarily aim to immediately improve safety by compensating for impairments (compensatory strategies) [41].

Motor learning interventions for infant feeding and swallowing typically aim to improve suck-swallow-breath coordination, oral skills for bolus formation or control, and swallowing skills while minimizing safety risks [41,50-53]. These interventions include biting or chewing practice with transitional foods which quickly

dissolve to puree, increasing oral awareness by altering taste, temperature or tactile quality of food, or refining bolus control and swallow through small controlled sips of water which has minimal chance of causing infection if aspirated [41,54].

The application of these interventions must follow some principles: (1) Use it or lose it (feeding skills may plateau or diminish if infant receives no or limited opportunity to utilize oropharyngeal skills for feeding and / or oral exploration); (2) Timing (begin intervention as early as possible); (3) Maximize opportunity for practice (repetition is essential for skill acquisition); (4) Specificity (practice task as close to meal as possible); (5) Rehearsal strategy: simplification (compensatory strategies can assist in motor learning if they are used to simplify the task in early stages and are regularly titrated down to work towards performing the task unaided); (6) Attention and motivation (ensuring attention, enjoyment and motivation during snacks and at meals with preferred flavors and praise from carers); (7) Extrinsic feedback (knowledge of performance, remaining time or mouthfuls and praise provided frequently during early learning and intermittently as skill is mastered); (8) Implicit learning (natural positive reinforcement and feedback provided through enjoyment of food flavors); (9) Blocked practice (practicing desired skill with one food or drink); (10) Random practice (alternate fluid, chewable and non-chewable textures with each mouthful); (11) Distributed practice (practice skill during many short sessions over a longer period); (12) Complexity (continual reassessment, upgrade goals and reducing support to ensure task is as difficult as possible, while maintaining safety); (13) Transfer of learning (once skill is mastered with familiar foods in familiar environments, skills can be practiced with novel foods and environments to encourage generalization of skills) [41,54-58].

Compensatory strategies are those which compensate for impairments by simplifying tasks, adding additional supports or modifying the environment to achieve success and immediately optimize safety. Among the compensatory strategies are modifying positioning to maximize control of muscles for feeding, utilizing alternative equipment such as slow flow nipples, adaptive equipment such as flexible cut-out cups, or thickening fluid consistency. Alternative and adaptive equipment limit flow rate and volume of fluid delivered, which allows additional time to control the bolus and coordinate swallowing, thus may minimize aspiration risk. Pacing is another compensatory strategy where the feeder periodically interrupts the feed to slow the rate or encourage breathing when the infant does not coordinate suck-swallow-breath patterns. Modifying texture of food to a moist, cohesive consistency is frequently recommended for people with poor oral control, as it requires less chewing or oral manipulation and reduces choking risk. Due to the serious consequences of dysphagia, compensatory strategies are often necessary and clinically indicated, as they immediately reduce risk of aspiration [41,59].

Approximatelly, in each group of 15 children with CP one will require non-oral feeding [60]. The indication of enteral tube-feeding regimens must be analyzed within a broad context of health, cultural, psychosocial and economic issues. In general, gastrostomy is indicated in the following situations: the caloric intake is insufficient to maintain growth; and / or there is a high risk of aspiration; and / or the level of effort required by the child and his caregiver is excessive to maintain an adequate caloric intake by the oral route [61]. In individuals with medically refractory GERD, fundoplication may also be used. In general, gastrostomy should be performed without empirical fundoplication in the absence of severe reflux [62-65]. Gastrostomy consistently leads to weight gain, may improve other growth measures including height and skinfold thickness [62], and leads to an improvement in QoL by reducing the frequency of aspiration pneumonia and hospitalizations [66,67]. The degree to which adverse events outweigh those benefits is likely to depend on the starting point of the individual, family stressors, and the degree to which adverse events can be mitigated using appropriate feed and other approaches [68]. Among the possible adverse events are the following: peritonitis, GERD, minor site infection, granulation tissue, leakage, tube blockages, tube migration, child pulling tube out, pneumonia, gastrointestinal bleeding and ulceration, wound dehiscence, and death [62,68-71]. Retrospective studies have suggested that gastrostomy may be correlated with higher than expected rates of mortality. In general, reported death rates range from 7% to 29%, with varying follow-up times. There are no data to suggest whether these rates are reduced relative to what would have occurred without treatment [62].

Current best available evidence for the treatment of dysphagia in CP suggests that a combination of motor learning interventions and compensatory strategies may lead to improved oral and pharyngeal function during feeding, feeding efficiency, decreased aspiration, increased oral intake and reduced reliance on tube-feeding. Clinicians should, therefore, monitor effects of chosen interventions using outcome measures and frequently reevaluate the potential for reducing compensatory strategies [41,72,73].

CONCLUSION

Dysphagia can negatively impact many dimensions of the patient's health, and the QoL of the patient and family. The highest morbidity and mortality in CP is related to respiratory impairment due to OPA, being dysphagia the main risk factor. Dysphagia and OPA influence each other through a vicious cycle worsening of swallowing / breathing coordination / aspiration axis. Early recognition and adequate management of dysphagia are very important in preventing CLD and premature death. The prevalence and severity of dysphagia are higher in cases of greater motor involvement. Dysphagia should be suspected and assessed in CP in all children with criteria based on the screening tool for feeding / swallowing difficulties and undernutrition [34], and / or those with GMFCS levels III, IV and V and / or those with red flags for dysphagia in order to improve swallowing skills, growth and nutritional outcomes and respiratory health. The interdisciplinary participation of medical specialties and rehabilitation professionals is essential for the assessment and treatment of intervening comorbidities. For the indication of any treatment, one should take into account, in addition to health issues, the socioeconomic and cultural characteristics of each family.

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