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Clinical signs suggestive of pharyngeal dysphagia in preschool children with cerebral palsy☆

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ABSTRACT

This study aimed to determine the discriminative validity, reproducibility, and prevalence of clinical signs suggestive of pharyngeal dysphagia according to gross motor function in children with cerebral palsy (CP). It was a cross-sectional population-based study of 130 children diagnosed with CP at 18–36 months (mean = 27.4, 81 males) and 40 children with typical development (TD, mean = 26.2, 18 males). Sixteen signs suggestive of pharyngeal phase impairment were directly observed in a videoed mealtime by a speech pathologist, and reported by parents on a questionnaire. Gross motor function was classified using the Gross Motor Function Classification System. The study found that 67.7% of children had clinical signs, and this increased with poorer gross motor function (OR = 1.7, $p < 0.01$). Parents reported clinical signs in 46.2% of children, with 60% agreement with direct clinical mealtime assessment (kappa = 0.2, $p < 0.01$). The most common signs on direct assessment were coughing (44.7%), multiple swallows (25.2%), gurgly voice (20.3%), wet breathing (18.7%) and gagging (11.4%). 37.5% of children with TD had clinical signs, mostly observed on fluids. Dysphagia cut-points were modified to exclude a single cough on fluids, with a modified prevalence estimate proposed as 50.8%. Clinical signs suggestive of pharyngeal dysphagia are common in children with CP, even those with ambulatory CP. Parent-report on 16 specific signs remains a feasible screening method. While coughing was consistently identified by clinicians, it may not reflect children's regular performance, and was not sufficiently discriminative in children aged 18–36 months.

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Abbreviations: CP, cerebral palsy; CPFQ, Queensland Cerebral Palsy Feeding Questionnaire; GMFCS, gross motor function classification system; GNPA, Growth, Nutrition, and Physical Activity (study); NHMRC, National Health and Medical Research Council (Australia); OPD, oropharyngeal dysphagia; SD, standard deviation; TD, typical development; VFSS, Videofluoroscopic Swallow Study.

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1. Introduction

Oropharyngeal aspiration (food or fluid entering the trachea below the vocal folds) (Brockett, 2006) is a commonly cited risk factor for recurrent pneumonia (Vaughan & Katkin, 2002) occurring frequently in children with non-ambulatory cerebral palsy (CP) and oropharyngeal dysphagia (OPD) (Mirrett, Riski, Glascott, & Johnson, 1994). In addition to causing pneumonia, chronic aspiration may lead to interstitial lung disease, pulmonary fibrosis and bronchiectasis (Lefton-Greif & McGrath-Morrow, 2007; Vaughan & Katkin, 2002). The nature of the aspirate, amount and frequency of aspiration all influence the consequent health outcomes, although the progression of respiratory sequelae and prognosis in children with CP are poorly understood (Cass, Wallis, Ryan, Reilly, & McHugh, 2005; Lefton-Greif & McGrath-Morrow, 2007). Factors related to respiratory status are of utmost importance, as respiratory-related factors are a leading cause of premature mortality in individuals with CP (Blair, Watson, Badawi, & Stanley, 2001). CP is a motor disability arising from a non-progressive neurological lesion, impacting on the strength and coordination of motor control (Smithers-Sheedy et al., 2013). As such, neurologically mediated mechanisms can compromise the sensorimotor tasks of eating and drinking; with impairments (OPD) occurring at any of the four phases of swallowing, including the oral-preparatory, oral-propulsive, pharyngeal and oesophageal phases (Matsuo & Palmer, 2008).

The pharyngeal phase involves a complex set of sensory and motor responses as food or fluid pass through the pharynx (Matsuo & Palmer, 2008). The pharynx is a shared anatomical juncture, involved in the functions of swallowing and respiration; hence, airway protection to prevent aspiration before, during and after bolus passage through the pharynx is critical for respiratory health. Airway protection is achieved by closure of the true and false vocal folds, the epiglottis inverting in response to the hyo-laryngeal excursion during the swallow, and finally deglutitive expiratory airflow (glottal release) (Lefton-Greif & McGrath-Morrow, 2007). Pharyngeal phase impairments in children with neurological conditions include inadequate airway protection during the swallow, incomplete laryngeal clearance following the initial swallow efforts, and/or decreased strength of pharyngeal contraction resulting in persistent residue in the hypopharynx post-swallow (Morton, Minford, Ellis, & Pinnington, 2002). Problems with the volitional oral motor movements of the oral-preparatory and propulsive phases may also affect bolus transit and thus compromise airway protection.

In addition to the specific neurophysiological limitations to the oropharyngeal mechanism, OPD in children with CP is also associated with their gross motor function (Benfer et al., 2013; Calis et al., 2008; Fung et al., 2002; Parkes, Hill, Plat, & Donnelly, 2010; Reilly, Skuse, & Poblete, 1996; Sullivan et al., 2000; Waterman, Koltai, Downey, & Cacace, 1992). An unstable pelvis and trunk can result in poor head and neck positioning, reducing the ability for controlled oropharyngeal movements (Bosma, 1992; Langley & Thomas, 1991). Children with CP may use disordered patterns of movement to create a base of stability, such as scapular retraction, which can influence the position of the oropharyngeal structures and restrict their mobility (Arvedson, Brodsky, & Reigstad, 2002). Poor head position has also been related to compromised airway protection by opening the airway, and the influence of gravity on flow rate of foods/fluids swallowed (Arvedson et al., 2002; Ekberg, 1986; Lanert & Ekberg, 1995).

The safety of the swallow is initially screened for clinically, including a comprehensive evaluation of the mealtime, and observation of clinical signs suggestive of pharyngeal phase impairment. A number of clinical signs have been used to indicate aspiration, with varying levels of sensitivity/specificity when compared to instrumental assessment, depending on the texture being assessed (Arvedson, Rogers, Buck, Smart, & Msall, 1994; DeMatteo, Matovich, & Hjartarson, 2005; Rogers, Arvedson, Buck, Smart, & Msall, 1994; Warms & Richards, 2000; Weir, McMahon, Barry, Masters, & Chang, 2009). When food or fluid reaches the vocal folds, a protective cough may be triggered, although children with CP are at high risk of 'silent aspiration' (no coughing when foods/fluids are aspirated), reported in between 82% (Weir, McMahon, Taylor, & Chang, 2011) and 94% (Arvedson et al., 1994) of cases of aspiration. It is therefore important in this population to observe other signs of aspiration such as wet/gurgly respiration or phonation, and fremitus (rattly chest). A child with clinical indications of aspiration may have this confirmed through evaluation with videofluoroscopic swallow study (VFSS). While widely considered the gold standard for detecting aspiration, VFSS tends to be restricted to tertiary hospitals (requiring trained personnel) and children are exposed to radiation during the procedure. Thus referral rates have remained relatively low, depending on the geographical region (Clancy & Hustad, 2011; DeMatteo et al., 2005; Waterman et al., 1992).

A number of studies have explored the patterns of pharyngeal phase impairments in CP, using clinical (Arvedson et al., 1994; Calis et al., 2008; Dahl, Thommessen, Rasmussen, & Selberg, 1996; Del Giudice et al., 1999; Erkin, Culha, Ozel, & Kirbiyik, 2010; Fung et al., 2002; Gerek & Ciyiltepe, 2005; Reilly & Skuse, 1992; Reilly et al., 1996; Rogers et al., 1994; Santoro et al., 2012; Sullivan et al., 2000; Wilson & Hustad, 2009; Yilmaz, Basar, & Gisel, 2004) and instrumental assessments (Arvedson et al., 1994; Field, Garland, & Williams, 2003; Gisel, Applegate-Ferrante, Bensen, & Bosma, 1995; Griggs, Jones, & Lee, 1989; Helfrich-Miller, Rector, & Straka, 1986; Morton et al., 2002; Rogers et al., 1994; Waterman et al., 1992; Weir et al., 2007, 2011; Wright, Wright, & Carson, 1996), but estimates of specific clinical signs of pharyngeal phase impairment have varied significantly. Many of the studies identified clinical signs through parent-report and only recruited children with moderate-severe CP or those with OPD. Further, children were either school-aged or recruitment spanned a broad age range (from infancy to adolescence). Coughing and/or choking (17–100%) (Del Giudice et al., 1999; Gerek & Ciyiltepe, 2005), gagging (14–69%) (Rogers et al., 1994) (Wilson & Hustad, 2009), and regurgitation (2.5–45%) (Erkin et al., 2010) (Reilly et al., 1996) were most frequently reported. There is generally consensus from instrumental assessment that thin fluids are the most likely food/fluid consistency to be aspirated in children with CP (Arvedson et al., 1994; Gisel et al., 1995; Morton et al., 2002; Rogers et al., 1994; Weir et al., 2007, 2011).