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삼킴장애를 가진 유아 및 아동의 삼킴 시간 측정 : 비디오조영검사 예비연구

Temporal Characteristics of Swallowing in Infants and Young Children with Dysphagia : A Preliminary Videofluoroscopic Study

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Purpose : Swallowing disorders during infancy and childhood can be related to medical complications. Appropriate swallowing assessment leads to effective swallowing management and a reduced risk of medical complications. The purpose of this study was to examine the temporal characteristics of swallowing in infants and young children with dysphagia in order to understand swallowing physiology and pathophysiology. **Methods :** Videofluoroscopic swallowing examinations (VFSEs) were obtained from 10 children who showed penetration or aspiration. Penetration was defined as entry of bolus into laryngeal vestibule, not passing the true vocal folds. Aspiration was identified when the bolus passed the true vocal folds. Each subject was fed a thin liquid by bottle or spoon. Five temporal measurements were analyzed, including oral filling, oral transit time, pharyngeal delay time, appearance of bolus to initiation of laryngeal closure, and pharyngeal transit time on the thin liquid. In addition, the five measurements were compared to those of a normal control group (Weckmueller et al., 2011). Descriptive analysis was used to compare group means in temporal measurements. **Results :** The oral filling, pharyngeal delay time, and pharyngeal transit time lasted longer for infants and young children with dysphagia than for infants and young children with normal swallowing. **Conclusions :** Infants and children who demonstrated specific oral and pharyngeal swallowing pathophysiology may have a high risk of penetration or aspiration. Normative data could serve as a reference to monitor changes in swallowing function of pediatric populations over time. Thus, future research is needed to analyze various temporal measurements to understand swallowing physiology in this population.

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목적 : 유아 및 아동기의 삼킴장애는 다양한 의학적 합병증을 야기할 수 있다. 유아 및 아동의 삼킴기능에 대한 이해는 적절한 삼킴평가와 효과적인 삼킴치료를 실행할 수 있게 하며 유아와 아동의 성장에 위험한 합병증을 줄일 수 있을 것이다. **방법 :** 비디오투시조영검사 중 침습이나 흡인을 보인 10명의 유아 및 아동의 삼킴을 분석하였다. 구강채움시간, 구강통과시간, 인두삼킴 지연시간, 음식덩이 출현부터 후두폐쇄까지 걸리는 시간, 인두통과시간을 측정하였다. 침습이나 흡인을 보인 유아 및 아동의 시간 측정 결과를 삼킴장애가 없는 정상유아 및 아동의 시간 측정 결과 (Weckmueller et al., 2011)와 비교하였다. 두 집단의 시간 측정 결과를 비교하기 위해서 기술적 분석을 실시하였다. **결과 :** 침습이나 흡인을 보인 유아 및 아동의 구강채움시간, 인두삼킴 지연시간, 음식덩이 출현부터 후두폐쇄까지 걸리는 시간, 인두통과 시간이 정상 대조군의 시간 측정값보다 길었다. **결론 :** 본 연구에서 구강과 인두에서 특정한 삼킴 병리적 현상을 보이는 유아 및 아동이 침습이나 흡인을 보일 위험성이 높은 것으로 나타났다.

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Ⅰ. Introduction

The incidence of feeding and swallowing disorder has increased due to an improvement of survival rate of children with history of prematurity and life-threatening medical disorders (Arvedson, 2008; Lefton-Grief, 2008). Feeding and swallowing disorders during infancy and early childhood can lead to clinical compromises, such as malnutrition, recurrent pneumonia, chronic lung disease and stunted growth (Arvedson, 2008; Burklow et al., 1998; Newman et al., 2001).

Multiple factors contribute to swallowing disorders in pediatric populations, including history of prematurity, cerebral palsy, congenital/acquired brain injury, neuromuscular disorders, respiratory disease, congenital cardiac disease, craniofacial anomalies, airway malformations, and gastrointestinal disease (Dodrill & Gosa, 2015; Friedman & Frazier, 2000; Lefton-Grief & Arvedson 2008; Newman et al., 2001; Roden & Altman, 2013). Previous researches reported that children with medical disorders experienced various swallowing problems, including penetration, aspiration, bolus spillover while sucking, weak suck, uncoordinated suck, delayed pharyngeal response, delayed laryngeal closure, and pharyngeal residue (Arvedson et al., 1994; Dodrill & Gosa, 2015; Newman et al., 2001). In addition, researchers noted that aspiration can be related to acute pneumonia or chronic lung disease in infants and children with dysphagia (Arvedson et al., 1994; Tutor & Gosa, 2012).

Swallowing can be divided into three stages; oral, pharyngeal, and esophageal. Each stage of swallowing has its own physiology events. In adults, physiological events were investigated using temporal and biomechanical measurements based on occurrence of penetration and aspiration. For example, delayed and/or reduced airway protection are directly related to penetration and aspiration (Park et al., 2010). The physiological events during swallowing provided important clinical decision-making process in diagnosis and treatment of swallowing disorders. There is limited investigation of temporal characteristics of swallowing in pediatric populations. Swallowing disorders in pediatric populations are different from those in adults. Diagnosis and treatment should target methods to reduce risk of aspiration and occurrence of other clinical complications. However, the underlying swallowing physiology and pathophysiology in the pediatric population remains poorly understood. The videofluoroscopic swallowing examination (VFSE) is the primary instrumental examination of swallow function. The VFSE provides clinicians with information about anatomic abnormalities, coordination of oropharyngeal muscles during bolus transition, and the timing and physiology of aspiration (Arvedson et al., 1994; Logemann et al., 2000). Temporal measurement of VFSEs provides quantitative value to examine physiologic events such as bolus transition, delayed pharyngeal swallow, and laryngeal closure during swallowing. Also, the use of quantitative data can identify physiological differences between normal swallowing and abnormal swallowing (Daniels et al., 2006). Previous research reported that patients who aspirated bolus showed longer pharyngeal delay time (PDT, Kim, 2004), delayed initiation of laryngeal closure (ILC, Park et al., 2010), prolonged bolus transition (Park et al., 2013; Power et al., 2009), and longer duration of Upper Esophageal Sphincter (UES) opening (DUESO, Power et al., 2009) than the adults who had no previously reported significant swallowing disorder.

As infants and young children have different anatomical structures and an immature swallowing mechanism compared to an adult's, temporal measures for these populations need to be modified (Petcu & Sasaki, 1991; Weckmueller et al., 2011). Currently, there is very limited research examining temporal characteristics to understand swallowing physiology in these populations. The purposes of this preliminary study were to examine temporal characteristics of swallowing in children with dysphagia and to compare temporal characteristics of children with dysphagia to those of a normal population from a previous research (Weckmueller et al., 2011). The findings of this study can provide more knowledge and understanding about swallowing physiology and pathophysiology and will allow future researchers to design appropriate management of dysphagia for infants and children.

Ⅱ. Methods

1. Subjects

Fifteen infants and children who were referred to Videofluoroscopic Swallowing Examination (VFSE) at Seoul National University, Korea were included in this study. Among them, 10 children who penetrated or aspirated on at least one bolus were selected as penetration/aspiration

(PEN/ASP) group. Penetration was identified when the bolus entered laryngeal vestibule but not passed the true vocal folds. Aspiration was identified when the bolus passed the true vocal folds. This group included six males and four females aged 5 months to 9 1/2 years. These infants and children have the following disease: encephalopathy (e.g., cerebral palsy, stroke, meningoencephalitis), syndromes (e.g., Prader-Willi syndrome, Down syndrome), neuromuscular diseases (e.g., myotonic dystrophy, mitochondria myopathy), or others (e.g., seizure disorder, gastointestinal disorder, delayed development) (Table 1). Data of the normal group (15 children) were obtained from a previous research as a control group (Weckmueller et al., 2011).

2. Procedures for temporal measurements

Each subject swallowed thin liquid. They were fed by bottle or spoon. First, the author examined the presence of penetration or aspiration for thin liquid swallows. Thin liquid bolus was mixture of milk formula or water and barium sulfate powder (35%w/v). The first five swallows of the subjects were submitted for analysis for the bottle feeding. For the spoon feeding, 2ml thin liquid bolus were submitted for analysis. Five temporal measurements were performed using 30 frames per second VFSE videos in all swallows. To analyze five temporal measurements, the author used slow motion frame-by-frame analysis by using Premiere Pro CS5.5 and a 100ms video timer. The five temporal measurements were as follows.

표 1. 연구대상 환자 정보 및 침습 또는 흡인 여부
Table 1. Demographic information of the subjects

Subject	Age /gender	Etiology	PEN or ASP[a]
1	1:8/M	Prader-Willi syndrome	PEN
2	8:1/M	Left middle cerebral artery infarction	PEN
3	1:5/F	Gastrointestinal disorder	PEN
4	1:5/F	Seizure disorder	ASP
5	0:6/F	Right thalamus, brainstem, bilateral cerebellar hemisphere infarction	PEN
6	4:2/M	Down syndrome	PEN
7	2:5/M	Hopoxic ischemic encephalopathy	PEN
8	9:6/M	Cerebral palsy	ASP
9	9:11/F	Acute inflammatory demyelinating polyneuropathy	PEN
10	0.5/M	Myotonic dystrophy	ASP

[a]PEN=penetration; ASP=aspiration.

1) Oral filling (OF)

The duration from the appearance of the bolus in the oral cavity until completion of oral filling (Weckmueller et al., 2011).

2) Oral transit time (OTT)

The duration in seconds between the initiation of posterior movement of the bolus head and the arrival of the bolus head at the ramus of the mandible (Power et al., 2009).

3) Pharyngeal delay time (PDT)

The duration in seconds between the bolus head passing the ramus of the mandible and the initiation of hyoid excursion (Logemann et al., 2000).

4) Bolus appearance to laryngeal closure (A to LC)

The time from bolus appearance in the oral cavity until first contact of arytenoids and epiglottis (Weckmueller et al., 2011).

5) Pharyngeal transit time (PTT)

The duration in seconds between the arrival of the bolus head at the ramus of the mandible and the bolus tail passing the upper esophageal sphincter (UES, Power et al., 2009). In addition to temporal measurements, physiologic swallowing components (e.g., lip closure, soft palate elevation, residue in oral cavity and pharyngeal cavity) were reviewed.

3. Reliability

The 20% swallows from the sample were reanalyzed by the author and a second judge for intra-judge and inter-judge reliability. The measurements of the first and the second judges were compared by using the Pearson correlation coefficient. The significant level of two reliabilities was designated at $p<.05$.

4. Data analysis

Descriptive analysis was used for comparison of group mean and standard deviation in temporal measurements. It was not possible to obtain raw data of the normal group. Therefore, the author used the normal population published mean and standard deviation data.

Ⅲ. Results

1. Reliability

The measurements for intra-judge and inter-judge reliability were compared by using the Pearson correlation coefficient. For intra-judge reliability, the author reanalyzed 6 swallows randomly selected (20% of swallows). A significant correlation was observed (r = 0.98 for OF, r = 0.98 for OTT, r = 0.98 for PDT, r = 0.96 for A to LC, r = 0.98 for PTT, p<.01). For inter-reliability, the same 20% swallows were analyzed by the second judge, an experienced graduate clinician. Significant correlations between the judges were observed (r = 0.98 for OF, r = 0.98 for OTT, r = 0.87 for PDT, r = 0.98 for A to LC, r = 0.98 for PTT, p<.01).

2. Temporal measurements

In the PEN/ASP group, three subjects showed aspiration and seven subjects showed penetration. In the oral phase, the PEN/ASP group showed longer OF (mean: 1.16 sec) and shorter OTT (mean: 0.62 sec) than the normal control group (OF: 0.69 sec; OTT: 0.89 sec) (see Figure 1).

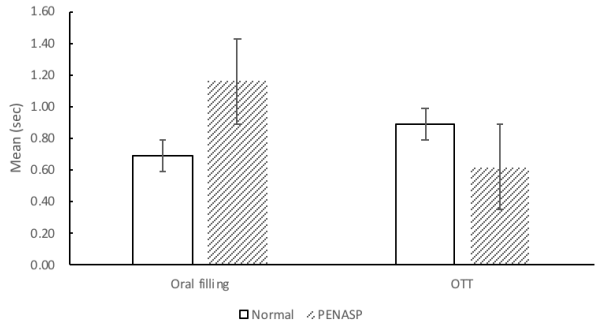

그림 1. 두 집단 간 구강채움시간 및 구강통과시간

Figure 1. Mean and standard deviation of oral filling and oral transit time for the two groups.

In the oropharyngeal transition phase, the PEN/ASP group had longer PDT (mean: 0.59 sec) and A to LC (mean: 1.41 sec) than the normal control group (PDT: -0.23 sec; A to LC: 1.04 sec) (see Figure 2).

In the pharyngeal phase, the PEN/ASP group showed longer PTT (mean: 0.63 sec) than the normal control group (mean: 0.39 sec) (see Figure 3).

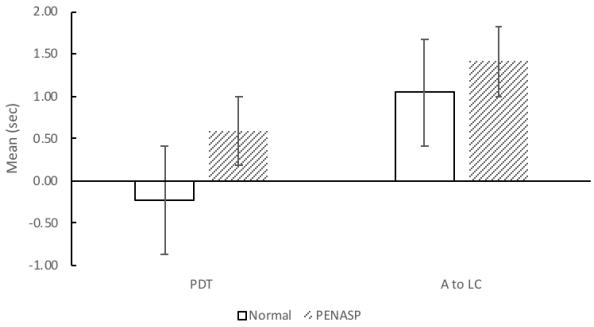

그림 2. 두 집단 간 인두삼킴 지연시간 및 음식덩이 출현부터 후두폐쇄까지 걸리는 시간

Figure 2. Mean and standard deviation of pharyngeal delay time and bolus appearance to laryngeal closure for the two groups.

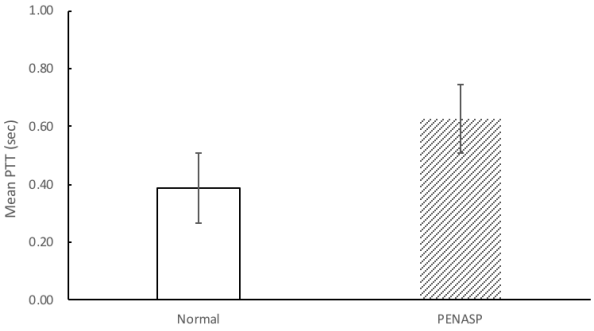

그림 3. 두 집단 간 인두통과시간

Figure 3. Mean and standard deviation of pharyngeal transit time for the two groups.

Ⅳ. Discussion

This preliminary study investigated the temporal characteristics of oropharyngeal swallowing in children with penetration or aspiration. Also, the temporal measurements of children with penetration or aspiration were compared to those of children without penetration or aspiration. Timing measurements of swallowing using VFSE will provide valuable information about swallowing physiology and pathophysiology in infants and children. The results of this study may be a good start in the identification of specific temporal measures that can distinguish children with dysphagia from children without dysphagia.

An inter-related pattern of disorders in oral phase was observed in this study. Some infants and children showed both longer OF and longer OTT as compared to the others in the PEN/ASP group. In addition, these children showed oral residue, excessive tongue pumping, or lack of efficient sucking. Previous research reported that

children with developmental disabilities have difficulty during the oral phase of swallowing, including absent oral reflexes, weak suck, and poor bolus propulsion (Arvedson 2008; Dodrill & Gosa, 2015). The PEN/ASP group had longer OF than the normal control group. Newman et al. (1991) identified that bolus transition time in oral phase increased as the number of sucks increased. The result of this study may indicate that children with dysphagia tend to have inefficient sucking. OTT was overall shorter in the PEN/ASP group than in the control group. In bottle-swallows, the valleculae contained the bolus while the subject was sucking (Newman et al., 1991). Two children in the PEN/ASP group did not collect the bolus in valleculae at all and the bolus entered the pharynx directly. In terms of temporal measurements, two children had relatively shorter OTT and longer PDT. Thus, excessively short OTT with delayed pharyngeal swallow may be related to a higher risk of aspiration.

Newman et al. (2001) reported that the most common cause of penetration and/or aspiration was delayed pharyngeal response. Henderson et al. (2016) reported that children showing arrival of the bolus at the pyriform sinus before airway closure had a higher score on penetration-aspiration scale. In addition to delayed pharyngeal swallow, impaired pharyngeal sensitivity can contribute to penetration or aspiration (Ekberg & Nylander, 1982; Friedman & Frazier, 2000). The PEN/ASP group in this study showed longer PDT and longer A to LC than the normal control group. These findings indicated that delayed hyolaryngeal excursion and delayed laryngeal closure is related to penetration and aspiration in pediatric populations. Longer A to LC in the PEN/ASP group may indicate that penetration or aspiration is due to delayed laryngeal closure. However, since A to LC included the measurement of oral filling, a future study needs to consider measurements of laryngeal closure, such as initiation of laryngeal closure and laryngeal closure duration, to examine laryngeal closure in pediatric populations.

The PEN/ASP group showed longer PTT than the normal control group. This seems to indicate that children with penetration or aspiration may have a weaker pharyngeal propulsion. Henderson et al. (2016) identified a positive correlation between weak pharyngeal constriction and longer PTT in children with dysphagia. According to Arvedson et al. (1994), children with disordered pharyngeal motility showed aspiration during VFSE. In this study, three children with aspiration were considered as outliers due to having longer PTT. They showed pharyngeal residue in valleculae and/or pyriform sinus more than the children showing only penetration.

The preliminary findings of temporal measures revealed that delayed pharyngeal swallow can be associated to potential penetration or aspiration. Position and posture changes can be a possible strategy for those who showed delayed pharyngeal swallow. For the younger children, postural change during feeding may be good strategy. Leaning toward stronger side will help the children transport the bolus safely in the pharynx. Larnert & Ekberg (1995) identified that chin-tuck with 30-degree reclined position reduced the occurrence of aspiration in children with cerebral palsy. A widened valleculae by chin-tuck held the bolus for a period of time so that the time for initiation of pharyngeal swallow increased. Other swallowing maneuvers, including thermal stimulation, supraglottic and effortful swallow, may help older children facilitate the swallow response and laryngeal closure (Lefton-Grief, 2008; O'Donoghue & Bagnall, 1999).

This study had several limitations, including the small number of subjects, which required a descriptive analysis which provide lacking information to compare temporal measurements between the groups. The range of subject's age are relatively big. This data may not represent the population of pediatric dysphagia population but will be a good foundation for the future study. Additionally, the published data were used for the normal group, resulting in a limited number of temporal measurements available for analysis. The author contacted the previous researchers to obtain normal data of temporal measurements, but they could not share them with other institute. Thus, future studies should recruit more subjects to conduct inferential statistical analysis. Also, additional research regarding temporal measurements that examine laryngeal closure and function of the upper esophageal sphincter during oropharyngeal swallowing would provide clinicians with more information to understand the swallowing function in pediatric populations.

This study is part of a long-term project, which will collect more normative data about swallowing physiology in pediatric populations. Normative data could serve as a reference to monitor changes in swallowing function of pediatric populations over time. It is important to note that a swallowing evaluation needs to identify types and physiological causes of swallowing disorders. It is crucial for valid and reliable temporal measures to be developed so that normal swallowing is more easily distinguished from abnormal swallowing. Additionally, these temporal measure developments will assist with determining the

relationship between swallowing physiology events and aspiration. Overall, the valid and reliable temporal measures determined by current and future studies will provide clinicians with evidence to facilitate appropriate intervention.

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