Critical Review: The Cause of Drooling in Children with Cerebral Palsy: Hypersalivation versus Swallowing Impairment.

Henderson, K.
M.Cl.Sc. (SLP) Candidate
School of Communication Sciences and Disorders, U.W.O.

This critical review examines hypersalivation versus swallowing impairment as a possible cause of drooling in children with cerebral palsy. Studies using a correlational case-control design were analyzed. Overall, the literature does not support hypersalivation as a cause of drooling in children with cerebral palsy.

Introduction

Cerebral palsy refers to a group of chronic, non-progressive disorders of movement, posture, and tone due to central nervous system damage (Marieb et al., 2005). This damage typically occurs prenatally, perinatally, or in the first 3 years of life (Ceriati et al., 2006). Children with cerebral palsy have a wide range of characteristics and may experience speech difficulties and other motor impairments such as swallowing disorders and oral motor dysfunctions.

Some children with cerebral palsy have the difficulty with sialorrhea or drooling. Drooling is defined as the spilling of saliva from the mouth onto the lips, chin, neck, and clothing (Senner et al., 2004). Drooling typically occurs in infants and young children, particularly when a child is learning a new motor skill or cutting a new tooth. Typically developing children around the age of 24 months old should have the ability to perform most activities without drooling (Senner et al., 2004). However, abnormal drooling primarily results from dysfunctional voluntary oral motor activity, improper swallowing, oral sphincter deficits, and or hypersalivation (Sochanjwskyj et al., 1986).

It is estimated that drooling abnormally persists in 10 to 38 percent of individuals with cerebral palsy causing both medical and social consequences (Senner el, 2004; Tahmassebi & Curzon, 2003). The medical consequences of drooling include irritated facial skin, unpleasant odor, increased oral and perioral infections, problems with hygiene, and dehydration. In addition to the medical consequences of drooling, there exist many social implications; the most devastating being social isolation. Drooling is unsightly and produces an unpleasant odor, causing individuals to reduce physical contact and avoid individuals who drool. This social isolation can have devastating effects on an individual's self-esteem (Senner et al., 2004). Since drooling affects a great number of children with cerebral palsy the question becomes, why are these children prone to drooling problems?

One side of the debate defines the cause of drooling as hypersalivation, or the excessive production of saliva. In a healthy individual, the average saliva flow rate is about 0.3 ml per minute and serves many functions (Ceriati, et al., 2006). It is thought that children with cerebral palsy produce more saliva than typically developing children, and therefore, the excess saliva is believed to be the cause of drooling.

The other side of the debate proposes that children with cerebral palsy drool due to swallowing dysfunction. Swallowing is a highly complex act which incorporates sequential and patterned movements of the lips, tongue, palate, jaw, pharynx, larynx and respiratory muscles. Swallowing is generally divided into three phases: oral phase, pharyngeal phase, and esophageal phase (Sochaniwskyj et al., 1986). It is thought that children with cerebral palsy show similar swallowing patterns to typically developing children in the pharyngeal and esophageal stages of swallowing; however, children with cerebral palsy have difficulty in the oral stage of swallowing (Sochaniwskyj et al., 1986). This difficulty in the oral stage of swallowing is thought to be the cause of drooling in children with cerebral palsy.

Objectives

The objective of this review is to critically examine the literature to determine if drooling in children with cerebral palsy is caused by hypersalivation or swallowing impairment.

Methods

Search Strategy
Computerized databases, including CINAHL, PubMed, and Medline were searched using the following search strategy:

((cerebral palsy) AND (drooling OR sialorrhea) AND (hypersalivation) OR (swallowing impairment) OR (swallowing dysfunction))

The search was limited to articles written in English between 1980 and 2006.

Selection Criteria
Studies selected for inclusion in this critical review paper were required to investigate whether drooling in children with cerebral palsy was caused by hypersalivation or swallowing impairment.

Data Collection
Results of the literature search yielded four correlational case-control studies.

Results

Senner et al. (2004) describe a correlational case-control design of 42 participants, 14 participants with cerebral palsy who drooled, 14 participants with cerebral palsy who did not drool, and 14 control participants. The study looked at whether drooling was caused by hypersalivation versus swallowing impairment using the Saxson test of whole saliva collection, cervical auscultation and videotaping to measure swallowing function. Results were analyzed using a one-way analysis of variance (ANOVA). The authors reported significant results. The results were further analyzed using a Tukey/Kramer post hoc analysis. Results of the study suggest children with cerebral palsy who drool have more severe oral motor involvement and a tendency to swallow less than children with cerebral palsy who do not drool. In addition, participants with cerebral palsy did not produce excess saliva, suggesting that hypersalivation is not one of the factors responsible for drooling in children with cerebral palsy.

Tahmassebi and Curzon (2003) describe a correlational case-control design of 20 participants, 10 participants with cerebral palsy who drooled and 10 participants control participants. The study looked at whether drooling was caused by hypersalivation versus swallowing impairment using the chin-cup method of drool collection. Results were analyzed using a two sample (unpaired) *t* test and a Fisher's exact probability calculation. The authors reported insignificant results. Results of the study suggest that there was no significant difference between salivary flow rates for the two groups. Therefore, hypersalivation was not a cause of drooling in children with cerebral palsy.

Sochaniwskyj et al. (1986) describe a correlational case-control design of 36 participants, 12 participants with cerebral palsy who drooled, 12 participants with cerebral palsy who did not drool, and 12 control participants. The study looked at whether drooling was caused by hypersalivation versus swallowing impairment using the chin-cup method of saliva collection and EMG recordings. Results were analyzed using a least-squares linear regression. The authors reported insignificant results. Results of the study suggest a trend towards inefficient and infrequent swallowing as the cause of drooling in children with cerebral palsy, rather than hypersalivation.

Lespargot et al. (1993) describe a correlational case-control design of 30 participants, 10 participants with cerebral palsy who drooled, 10 participants with cerebral palsy who did not drool, and 10 control participants. The study looked at whether drooling was caused by hypersalivation versus swallowing impairment using the EMG and oral pressure transducers. It is unclear from the text what statistical analysis was used on the data, therefore, making it difficult to evaluate the accuracy and significance of the results. Results of the study implicate the oral stage of swallowing to be the cause of drooling in children with cerebral palsy.

Discussion

Participant Selection
A between group design was used in each study where children with cerebral palsy were compared to typically developing children. Three of the studies divided participants into three groups, children with cerebral palsy who drooled, children with cerebral palsy who did not drool, and a control group (Senner et al. 2004; Schoaniwskj et al. 1986; Lespargot et al. 1993). Tahmassebi and Curzon (2003) divided participants into two groups of 10: children with

cerebral palsy who drooled and a control group. This study did not take into consideration children with cerebral palsy who did not drool, which excludes valuable information when determining the cause of drooling in children with cerebral palsy.

Participant selection is important to ensure the key characteristics being examined are present and controlled for. Therefore, inclusion/exclusion criteria for participant selection should be available in each study. Senner et al. (2004) and Schoaniwskj et al. (1986) provided inclusion/exclusion criteria for each group, controlling for type of cerebral palsy in particular. In addition, Senner et al. (2004) controlled for severity of drooling in children with cerebral palsy. Tahmassebi and Curzon (2003) and Lespargot et al. (1993) did not provide inclusion/exclusion criteria or control for type of cerebral palsy or severity of drooling. This is problematic as participants of the studies may not provide an accurate representation of the population. Also, by not controlling for type of cerebral palsy or severity of drooling the studies are introducing a number confounding variables. This may impact the overall results of the study as significant results may not be found, or if significant results are found the cause may be difficult to tease apart from the different variables.

A potential confound of each of the four studies is purposive sampling. A non-probability sample is chosen when individuals considered most closely related to the issue being studied are selected for inclusion (Portney & Watkins, 2000). Due to this, the sample may not be representative of the population; however, due to the nature of the studies, purposive sampling is an adequate method of participant selection. In addition, a rationale was not provided for sample size in any of the four articles. This may have an impact on the power of studies; therefore, insignificant results may be the effect of a small sample size rather than no difference existing between the groups.

Methodology

Senner et al. (2004) provided clearly stated and specific protocols increasing the reproducibility of the study; however, a potential confound was noted when only one drooling sample was collected for each participant using the Saxson test. Senner et al. (2004) rationalized this decision using a Spearman's rho correlation demonstrating that the Saxson test has high test-retest reliability. As well, Schoaniwskj et al. (1986) provided clearly stated and specific protocols allowing the study to be reproduced; however, a potential confound was identified when chin-cup drool spillage was not measured. This directly impacts the accuracy of the amount of drool reported for each participant, as drooled saliva that did not enter the chin-cup was not included in the amount of drooled saliva reported.

Tahmassebi and Curzon (2003) did not provide adequate details for protocols, and therefore, the study is not reproducible. In addition, the protocol for the group with cerebral palsy differed from the protocol for the control group. For example, the control group was measured on whole saliva production for a 5 minute collection sample, whereas the group with cerebral palsy was measured on drooled saliva for a 15 minute collection sample. These protocols are problematic because hypersalivation cannot be quantified using drooled saliva collection. Rather, hypersalivation can only be quantified using a whole saliva collection method. As well, spillage of drooled saliva for the group with cerebral palsy was not quantified; therefore, measurement of amount of saliva drooled appears to be inaccurate.

Finally, Lespargot et al. (1993) did not provide adequate details of protocols; making it very difficult to reproduce this study. Due to this, the results of the study cannot be reproduced and verified for accuracy.

Measurement tools and Outcome Measures

The studies conducted by Senner et al. (2004) and Sochaniwskj et al. (1986) provided operational definitions of outcome measures. The definitions were conventional and provided sufficient information to assume that measurement tools were accurately chosen. Tahmassebi and Curzon (2003) and Lespargot et al. (1993) did not provide definitions for all of the outcome measures. For example, Tahmassebi and Curzon (2003) measured the buffering capacity of saliva; however, did not provide a definition or a rationale as why measuring buffering capacity is beneficial.

Senner et al. (2004) demonstrated high test-retest reliability for the Saxson test, which is a technique used to collect whole saliva samples from the participants. In addition, Senner et al.

(2004) demonstrated high interrater reliability for standardized tests, and high interoberserver reliability for videotaping. Providing information on reliability of the above measures demonstrates their dependability. However, Senner et al. (2004) did not speak to the validity of the measures, the extent to which an instrument measures what it is intended to measure. Using the operational definitions, it can be concluded that all measurement tools were used for their intended purpose.

The remaining three studies did not discuss the reliability and validity of the measurement tools (Sochanjwski et al. 1986; Tahmassebi & Curzon, 2003; Lespargot et al. 1993). The operational definitions of Sochanjwski et al. (1986) allow for interpretation of the reliability and validity of the study. The tools used in the study appeared to have face validity.

The outcome measures not defined in Tahmassebi and Curzon (2003) study included loss of drooling, whole saliva, and buffering capacity of saliva. The definition used for the chin-cup method of drool collection was accurately defined; however, the purpose of the chin-cup is to measure drooled saliva when the study set out to look at hypersalivation. Hypersalivation can only be identified using a measurement of whole saliva production; therefore, demonstrating low measurement validity for identifying hypersalivation. Due to the lack of operational definitions provided by Tahmassebi and Curzon (2003), it is difficult to determine the validity of the measures.

Lespargot et al. (1993) did not include definitions for severity of drooling, collection of drool, length of trials, and residual residue in the oral cavity. By not providing definitions for all outcome measures, it is difficult to determine if all measurement tools were accurately used, therefore, reliability and validity could not be determined for this study.

Statistical Analysis

The statistical analysis of the data used by Senner et al. (2004) included the Spearman's rho correlation, the one way analysis of variance (ANOVA), and Tukey/Kramer. The Spearman's rho correlation was used to determine drooling severity as marked on a questionnaire. This non-parametric test was used to interpret ordinal data, which is an appropriate test. The ANOVA is a parametric test used with ratio data to look at the between group difference for the three independent groups (Portney & Watkins, 2000). The ANOVA was used to compare amount of saliva produced and drooled, swallowing frequency, and functional skills. All results were measure against $p < 0.05$. As multiple comparisons were made on a single sample of data the ANOVA is the acceptable test to use as it allows for multiple comparisons while maintaining a low type I error rate. In addition, the study further analyzed significant results found by the ANOVA with the Tukey/Kramer. This is an adequate post-hoc measure to determine which pairs of comparison accounted for the significant difference (Portney & Watkins, 2000).

The study conducted by Tahmassebi and Curzon (2003) analyzed data using a two sample (unpaired) *t* test and a Fisher's exact probability calculation. The two sample (unpaired) *t* test is a parametric test which was used with ratio data looking at the between group differences for salivary flow rates of the children with cerebral palsy and controls. As one comparison was made, the *t* test was the proper statistical tool as no further confounding results occurred (Portney & Watkins, 2000). The Fisher's exact probability calculation is a non parametric test which was used with ordinal data, comparing the buffering capacity of saliva for the two groups. This statistical analysis tool was used accurately as it determines if conditions are independent or associated.

Least-squares linear regression was used to analyze the data in the Sochanjwski et al. (1986) study. This parametric test was used on ratio data and was preformed to test the correlation between drooling rates and frequency of swallowing for the group with cerebral palsy who drooled. Least-squares linear regression was used accurately as it describes the numerical relation between two quantitative variables, allowing one value to be predicted from the other (Portney & Watkins, 2000).

Finally, the statistical analysis used in Lespargot et al. (1993) was unclear from the text. Therefore, it is hard to evaluate if the statistical analysis is accurate. The study should have used an ANOVA since ratio data was used to look at the between group differences for the three independent groups. In addition, an ANOVA allows multiple comparisons of data from a

single sample while keeping type I error rate low.

Recommendations

Review of the relevant literature indicates that drooling in children with cerebral palsy is not due to hypersalivation (Senner et al., 2004; Tahmassebi & Curzon, 2003; Sochanjwski et al., 1986). The literature further suggests swallowing impairment as a probable cause of drooling in children with cerebral palsy (Senner et al., 2004; Tahmassebi & Curzon, 2003; Sochanjwski et al., 1986; Lespargot et al., 1993). However, throughout the literature there was no concrete evidence to support swallowing alone. Rather, the studies suggest swallowing impairment coupled with oral motor dysfunctions as the cause of drooling in children with cerebral palsy.

Further research is required to precisely determine the cause of drooling in children with cerebral palsy. Future research should target the frequency as well as the different stages of swallowing in children with cerebral palsy. In addition, research should address oral motor functioning in children with cerebral palsy.

Conclusions

Hypersalivation is not a cause of drooling in children with cerebral palsy. Further research is required to determine if swallowing and oral motor function can be linked to drooling in children with cerebral palsy.

References

Ceriati, E., De Peppo, F., Ciprandi, G., Marchetti, P., Silveri, M., Rivosecchi, M., (2006). Surgery in disabled children: General gastroenterological aspects. Acta Paediatrica, (95) 452, 34-37.

Lespargot, A. Langevin, M. Muller, S. Guillemont, S. (1993). Swallowing disturbances associated with drooling in cerebral-palsied children. Developmental Medicine & Child Neurology, 35, 298-304.

Marieb, E. N., Mallatt, J., Wilhelm, P. B., (2005). Human anatomy. Fourth Edition. San Francisco, CA: Pearson Education, Inc.

Portney, L.G., & Watkins, M.P. (2000). *Foundations of Clinical Research: Applications to Practice* (2nd ed.). Upper Saddle River, New Jersey: Prentice Hall Health.

Senner, J. Logemann, J. Zecker, S. Gaebler-Spira, D. (2004). Drooling, saliva production, and swallowing in cerebral palsy. Developmental Medicine & Child Neurology, 46, 801-806.

Sochaniwskyj, A. E. Koheil, R. M. Bablich, K. Milner, M. Kenny, D. (1986). Oral motor functioning, frequency of swallowing and drooling in normal children and in children with cerebral palsy. *Archives of Physical Medicine and Rehabilitation*, 67, 866-874.

Tahmassebi, J. F. & Curzon, M. E. J. (2003). The cause of drooling in children with cerebral palsy – hypersalivation or swallowing defect? *International Journal of Paediatric Dentistry*, 13, 106-111.