Critical Review: The Effectiveness of Oral Sensorimotor Therapy in the Treatment of Drooling in Children with Cerebral Palsy

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This review examined the effectiveness of oral sensorimotor therapy in improving drooling in children with cerebral palsy. Seven studies were reviewed including single-subject studies, a randomized clinical trial, a randomized prospective study, and a case study. Therapy methods included the use of orthodontic appliances as well as various muscle training protocols. Oral sensorimotor therapy was determined to be effective in treating drooling in children with cerebral palsy. Speech pathologists should regard this therapy method as first line treatment because of its ability to reduce drooling while improving feeding skills such as chewing and swallowing without negative side effects.

Introduction

Cerebral palsy (CP) has been found to be the most common physical disability in early childhood (Erasmus et al., 2009). Rosenbaum and colleagues (2007) defined CP as a group of permanent disorders of the development of movement and posture that are attributed to nonprogressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of communication, cognition, sensation, perception, and behavior (Rosenbaum et al., 2007). Consequently, the speech musculature can be negatively affected, causing disruptions in intraoral tongue suction, lip closure, and swallowing (Erasmus et al., 2009). These motor issues combine to cause drooling (an important comorbidity) which is seen in approximately 40% of children with CP (Reid et al., 2012). Chronic drooling is a socially stigmatizing issue, it may cause a bad odour, lead to hygiene problems for caregivers and parents, and can interfere with interpersonal relationships (Thomas-Stonell & Greenberg, 1988).

A variety of treatments have been explored with the most common being: pharmacological, surgical, and botulinum toxin injections. Reviews conducted by Walshe, Smith and Pennington (2012) and Varley, Denieffe, O'Gorman, Murphy and Gooney (2019) have both come to the same conclusion: it is unclear which intervention is most effective and they all have their disadvantages. For example, botulinum injections have side effects including dry mouth, difficulty chewing and thickened secretions (Jackson et al., 2009; Varley et al., 2019). In terms of anticholinergic drugs, patients can suffer from various side effects including irritability and agitation (Mato et al., 2010). Surgery can also have complications including loss of tongue movement, hearing, and taste (Brei, 2003; Burton, 1991).

In addition to their downsides, the aforementioned treatment options do not address the root cause of drooling in individuals with CP. Results from a study conducted by Erasmus et al. (2009) support findings from previous studies that, in general, hypersalivation does not exist in CP. These authors suggested that dysfunctional oral motor control is what causes saliva overflow from the mouth (Erasmus et al., 2009). Reid et al. (2012) also found poor oromotor function to be associated with drooling in children with cerebral palsy and that it could be the target of intervention for this issue.

The present critical review explores the effect of oral sensorimotor therapy on drooling rates in children with CP. Sensorimotor interventions include using various methods to improve the degree of control over muscles, as well as their range and strength. This therapy also includes the use of intraoral appliances to accomplish similar goals.

Objectives

The primary objective of this paper is to evaluate the literature regarding the effectiveness of oral sensorimotor therapies as treatment for children with cerebral palsy who drool. The secondary objective is to propose a recommendation regarding the use of oral sensorimotor therapy for children with cerebral palsy who have chronic drooling.

Methods

Search Strategy

Computerized databases including PubMed, Google Scholar, and Medline were explored using the following search strategy:

> (drooling) OR (sialorrhea) AND (cerebral palsy) AND (treatment) OR (intervention) OR (therap*) OR (behav* mod*) OR (remedy)

The search was limited to articles written in English.

Selection Criteria

Studies included in this review were investigations into the impact of an oral sensorimotor therapy on reducing drooling rates in children with cerebral palsy.

Data Collection

Results of the literature search yielded the following study designs meeting selection criteria: Single-Subject studies (4), a Randomized Clinical Trial, a Randomized Prospective Study, and a Case Study.

Results

Single-Subject Studies:

Fischer-Brandies et al. (1987) assessed the effectiveness of using stimulatory plates in order to treat orofacial dysfunctions in children with cerebral palsy. These 71 children (mean age = 10 years) received this orofacial regulation therapy for an average of 1 year and 3 months. The removable plates inhibited abnormal tongue and lip positioning while facilitating normal movement of these structures. As the types of CP varied in the subjects, so too did the appliances. These variations included addressing the cigar-shaped spastic tongue and alternating borders to practice lateral tongue movement. In addition to treatment with these plates, oral and facial physiotherapy was applied in one third of the participants. The oral sensory-motor abilities of these children were assessed with a neuropaediatric exam at the beginning and end of treatment, with check-ups every 2-4 months. Outcome measures included various symptoms such as drooling, feeding issues, and tongue positioning and mobility. It is unclear if appropriate statistical analysis was conducted as they were not described. Results found improvement in at least one half of the participants in terms of drooling, spontaneous tongue position, tongue mobility, food intake, and speech development.

In addition to there being no control group, a major limitation of this study is that only 49 participants were treated with physiotherapy during the treatment process. This inconsistency renders it difficult to attribute the measured results solely to the orthodonic devices. The major strength of this study is the fact that pre and post assessments were conducted by the same neuropaediatrician using a specific and exact protocol which included common neurological examinations. These result in the study having both suggestive validity and importance.

Similarly, Johnson et al. (2004) evaluated the effectiveness of the Innsbruck Sensorimotor Activator and Regulator (ISMAR), an orthodontic appliance, in improving drooling and feeding skills in a group of children with cerebral palsy. This study ultimately included six children due to a high rate of withdrawal. The age of these participants ranged from 4-13 years. These children had motor disabilities including athetosis (n = 3), spastic quadriplegia (n = 2), and hypotonia (n = 1), with none having greater than mild cognitive impairment. During the initial assessment, baseline drooling severity was measured using the drooling rating scale. All participants had either moderate or severe levels of drooling. Dysphagia classifications were also determined at this time, and the participants had severity levels ranging from mild to severe. After assessment, the appliance was constructed after casts were taken and fittings were undergone. The devices included stimulators to accomplish a variety of goals depending on recommendations by the dentist and speech pathologist. These modifications included options to prevent unwanted movements (i.e., tongue thrusting) and those to encourage more complex tongue movements (i.e., lateral movements). The treatment phase of the study began when children were able to tolerate the ISMAR for a 10-minute session, which they were asked to do at least once a day. Once they were able to tolerate a 20-minute session, they were to wear it overnight. Participants were re-assessed at the beginning of the treatment phase and at the completion of the study. Outcome measures included frequency and severity of drooling, and various feeding skills such as chewing. Appropriate statistical analysis was performed and demonstrated significantly improved drooling severity scores for the six children who completed the study.

A limitation of this study is the fact that the children varied in the length of time they wore the appliances. It is also concerning that only 6 of 18 children were able to finish the study; it is clearly difficult for children with CP to comply with using this type of device. Ultimately, the authors concluded that the ISMAR is a valid treatment option for drooling in this population. This study has been determined to have suggestive validity and importance.

Koheil et al. (1987) assessed the efficacy of EMG auditory feedback in training the orbicularis oris, making the act of swallowing a conscious one, and providing an auditory signal to cue swallowing, all in order to decrease drooling rates. Participants included 12 children with cerebral palsy who drooled whose ages ranged from 6 to 18 years (M = 11.7 years). Assessment involved quantifying drooling rates and measuring swallowing frequency. Biofeedback training then took place by spending time practicing lip puckering, lip approximation, sucking, blowing, and swallowing. The children were to turn the auditory feedback on and off with appropriate muscle activity. During this phase, children were also taught to swallow in response to an auditory signal. This was usually set

to be every 40 seconds but was made shorter if necessary. Once a participant achieved an 80% success rate, they were then to proceed to the next phase which involved using the auditory signal at a regular environment (i.e., school) for a minimum of two hours a day. Participants were then re-assessed, had one month without using the timing device, and then were re-assessed a final time. Outcome measures included drooling rate (ml/hr) and swallowing rate (swallows/hr). Appropriate statistical analysis was performed, and results found that drooling rates were significantly reduced and swallowing rates had increased. The authors concluded that this study highlights the usefulness of auditory feedback in improving oral motor skills to treat drooling.

Although this study had a small sample size, it is not without its strengths. The children all received the same training and instructions, making this study very replicable. The participants were also assessed at various times allowing for both short term and longer term data. This study was determined to have highly suggestive validity and importance.

Limbrock et al. (1990) conducted a six-year study to assess the effectiveness of using orthodontic appliances to treat orofacial dysfunctions such as drooling and chewing in children with cerebral palsy. The participants included 68 children whose ages ranged from 6 months to 16 years. Children received treatment for various amounts of time ranging from 1 to 3-4 years. Frequent examinations before and during therapy were completed by a multidisciplinary team in order to create the orthodontic appliances. The prosthetics were modified to address the needs of each type of cerebral palsy: spasticity, athetosis, and hypotonia. These included stimulating palatal plates to reduce tongue thrusting, lateral pendulum stimulators to train lateral tongue movements, and bumpers to stimulate upper and lower lips. Outcome measures included drooling, swallowing, tongue mobility, and chewing. It is unclear if appropriate statistical analysis was conducted as it was not described. However, results found improvements in drooling rates in 67% of all participants, and an improvement rate of 72% in cases of severe drooling. Improvements were also seen in 81% of participants in terms of chewing, and 64% in terms of swallowing.

The authors mention that a limitation of this study is the lack of a control group. In addition to this is the fact that treatment times varied greatly and assessment details were not reported. In terms of strengths of the study, the authors go into great detail of the various modification options for each appliance. The authors also claim that the assessment criteria used in the study were reliable and that their clinical observations were replicable. However, the lack of detail in their assessment measures and process lead the validity of this paper to be equivocal. As the study found promising results and children were able to tolerate the treatment very well, the importance of the paper is highly suggestive.

Randomized Clinical Trial:

Gisel (1994) attempted to determine the effectiveness of oral sensorimotor treatment in improving eating skills in children with cerebral palsy. Participants included 35 children whose ages ranged from 4.3 to 13.3 years of age. The children's weight was at or above the 5th percentile for their age and all needed assistance with activities of daily living. Skinfold measures were taken and all participants were at or below the 35th percentile for their age. Following these tests, children were randomly assigned to one of three groups: 1) sensorimotor treatment 2) chewing-only treatment or 3) control. Groups 1 and 2 received 20 weeks of their treatments, and on weeks 10 and 20 were given the same tests as administered at the onset of the study. The Functional Feeding Assessment subtest (FFA) of the Multiple Feeding profile was used to assess the children's eating during a mealtime observation in their school's cafeteria. The participants were scored on 6 domains: spoon feeding, biting, chewing, cup drinking, straw drinking, and swallowing and drooling. Sensorimotor treatment was based on the child's individual needs and performance on the FFA. Emphasis was placed on three major areas: tongue lateralization, vigor of chewing, and lip control. Chewing-only treatment involved offering children small pieces of fruit gelatin to chew. Participants were given harder textures as they progressed, always being given 5-7 minutes to consume. Outcome measures included weight, skinfold measures, and eating-related behaviours such as eating-related drooling. Appropriate statistical analysis was used and results found eating-related drooling decreased from 63.6% in the sensorimotor treatment group to 45.5% after 20 weeks of treatment. No change in drooling was observed in the chewing-only condition or control group. It was also noted that children who received sensorimotor treatment gained more weight than those in the control group.

This paper appropriately assessed eating behaviours and weight of all participants at baseline, midpoint, and at completion. Treatment plans were also appropriate and properly described, ultimately giving this study compelling validity. This study clearly shows that sensorimotor treatment can reduce eating-related

drooling in children with cerebral palsy compared to control, therefore has highly suggestive importance.

Randomized Prospective Study:

Sigan et al. (2013) conducted a study to determine the effectiveness of oral motor therapy in children with cerebral palsy in improving problems with feeding. Participants were 81 children aged 12-42 months with cerebral palsy and oral motor dysfunction. These children had at least one or more issues associated with poor oral motor functions such as drooling, swallowing, chewing, sucking, and independent feeding. Participants were randomly sorted into either the training or control group. Before and after training, all participants were evaluated by various methods including the FFA and an oral motor assessment. In terms of treatment, the oral motor therapy involved 1-hour sessions with a physiotherapist once a week for 6 months. Tactile and proprioceptive aspects of eating were increased in order to improve swallowing and chewing. In order to improve mouth function and control, food textures were gradually thickened and families were taught proper positioning. Posture control, positioning, and mouth control were taught in order to reduce rates of drooling. Outcome measures included drooling and tongue and jaw functions. Appropriate statistical analysis was conducted and comparison between groups revealed the training group demonstrated a significant reduction in drooling compared to control. The training group also showed a significant improvement in average FFA scores as compared to the control group.

The authors concluded that oral motor therapy has a beneficial effect on the functioning of children with CP. The large sample size, strong methodology, and significant results lead this study to have compelling importance and validity.

Case Study:

Haberfellner & Rossiwall (1977) evaluated the effects of using an orthodontic appliance to treat the oral sensorimotor issues in a child with cerebral palsy. This child's tongue was consistently protruded and essentially immobile. He also exhibited mouth breathing, constant drooling, and a hyperactive gag reflex. The treatment device was an oral shield joined by wires to prosthetics in the inferior and superior labial vestibules. The oral shield covered the palate and extended downward inside the teeth to prevent abnormal tongue protrusion and induce a favourable resting tongue position. This appliance was tolerated within about three weeks and was used for increasing periods of time – from a few sessions of up to half an hour a day to nightly use. During the first year of therapy, lip closure became more normal and drooling diminished. After 18 months of therapy, no further drooling occurred and lip closure was adequate. At age 6.5 years, the child only used the appliance at night and exhibited normal lip closure, swallowing, and oral sensibility.

As this was a case study, results should not be generalized to all cerebral palsy patients who drool. Assessment of the child's anatomy and oral motor function seems to have been subjective and would have benefitted from the use of a standardized assessment protocol. However, the appliance and recommendations for use were well described making the study highly replicable. This leads the study to have somewhat suggestive validity but highly suggestive importance.

Discussion

The current available literature indicates that oral sensorimotor therapy is an effective treatment for drooling in children with cerebral palsy. A large advantage of this therapy option is its various types and their ability to be tailored to the needs of each child. For example, if a child is severely mentally handicapped, a clinician may choose to trial an orthodontic appliance rather than attempting to train proper tongue or lip movements. The selected appliance can then be properly modified to target the chosen therapy goals.

Chronic drooling is a salient and undesirable issue. The nature of drooling may lead parents to seek its treatment without ever considering that it is only one of many symptoms of poor oral motor functioning and control. A benefit of oral sensorimotor treatment of any variety discussed is its efficiency; it is likely that chewing and swallowing will improve in addition to drooling.

While oral sensorimotor treatment can have unexpected positive effects with no unexpected negative effects, the opposite is true for other treatment options. Botox and surgery can decrease drooling, but both are invasive and are accompanied by their respective risks and side effects. While less risky, pharmacological interventions still typically cause negative side effects. Although more risks are involved with these treatment options, they still warrant consideration after attempting sensorimotor methods. If a patient is unable to tolerate an orthodontic appliance or make gains in muscular training, clinicians should discuss botox or pharmacological options with parents/guardians. Corrective surgeries should be the last to be considered in a treatment plan to reduce drooling in children with CP.

One disadvantage of oral sensorimotor treatments is the length of treatment time necessary to achieve results. This time will of course vary but will likely take upwards of 6 months. Parents/guardians should know this and consider this time period when choosing a treatment option. If waiting is not an option, parents may want to consider the following recommendation. Children could receive botox injections or pharmacological interventions simultaneously with oral sensorimotor therapy. This secondary treatment option would ideally decrease drooling during the oral training period, and then be tapered off when no longer necessary.

Although results of oral sensorimotor therapy are encouraging, further investigation is warranted. Future research should focus on the following:
1) Expanding on the study conducted by Gisel (1994) by observing drooling independently of eating.
2) Comparing the effectiveness of orthodontic appliances to muscle training treatments.
3) Conducting longitudinal studies to determine long term effects of oral sensorimotor treatments.
4) Eventually determining if oral sensorimotor treatment can be effective in treating drooling in other neurologic conditions such as Parkinson's disease and Amyotrophic Lateral Sclerosis.

Conclusion

This review demonstrates that oral sensorimotor therapies are effective in treating chronic drooling in children with cerebral palsy. Clinicians should feel confident in exploring this treatment path regardless of CP type, especially before more dangerous options (i.e., surgery). Teams should also be able to recommend an appropriate treatment based on mental status and oral motor assessments. Future research should determine most effective methods, their long-term results, and their ability to generalize to other disorder areas.

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