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The Role of Speech Therapy in Sialorrhea Management and Quality of Life: A Retrospective Study

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ABSTRACT

Objective: The objective of this study is to determine the impact of an intensive and a less intensive speech therapy regimen on pediatric sialorrhea patient and caregiver quality of life (QoL) as described by drooling impact scales (DIS).

Methods: A retrospective chart review included all pediatric patients from a secretion management clinic. There were two outpatient speech therapy programs: intensive (4 sessions/week for 3 weeks) and less intensive (2 sessions/week for 3 months). Both regimens included similar interventions including neuromuscular electrical stimulation and facial taping. The primary outcome measure was reduction in caregiver reported DIS after completing their designated program. Children observed without therapy also obtained DIS assessments overtime to measure potential changes in QoL.

Results: There were 49 patients included in the study with a mean age of 7.5 years (SD = 4.6). The most common comorbidities were global developmental delay ($n = 47$; 96%), epilepsy ($n = 35$; 71%), and cerebral palsy ($n = 32$; 65%). After initial evaluation, 30 patients underwent speech therapy with significantly improved DIS scores compared to the 19 who had no therapy (43.4 vs. 54.5, $p = 0.03$). Of these 30, 16 (33%) underwent intensive therapy with mean DIS improving from 63.5 to 47.2 ($p = 0.006$). Fourteen (29%) completed the less intensive regimen with mean DIS improving from 51.9 to 39.1 ($p = 0.07$). There were 19 (39%) patients who underwent no therapy and mean DIS remained unchanged from 55.6 to 54.5 ($p = 0.86$).

Conclusion: Sialorrhea can drastically impact patients and their families. An intensive speech therapy program is associated with improved QoL as described by the DIS. Speech therapy should be considered as an effective treatment modality to improve outcomes for pediatric sialorrhea.

Level of Evidence: 2.

1 | Introduction

Sialorrhea, or excessive drooling, affects about 60% of pediatric patients with neurological impairments, such as cerebral palsy, epilepsy, or developmental delay [1, 2]. Ideal management is often an interdisciplinary challenge [1]. Pediatric sialorrhea can

lead to chronic aspiration, resulting in pneumonias, frequent hospitalizations, and declining pulmonary function [3]. In addition, it can significantly impact both patient and caregiver quality of life (QoL) [4, 5]. This impact can be estimated via the drooling impact scale (DIS), a 10-point validated questionnaire used to evaluate the impact of sialorrhea [6, 7]. This validated

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scale is a valuable tool used to measure the efficacy of interventions in managing sialorrhea.

Sialorrhea management ranges from conservative treatments such as speech therapy and postural adjustments to more invasive procedures like salivary gland excision [1]. Anticholinergic medications such as atropine sulfate, glycopyrrolate, and scopolamine may be used for management, but can have adverse side effects such as constipation, flushing, visual disturbance, and urinary retention [8, 9]. Botulinum toxin A (BT-A) is a neurotoxin often used to decrease saliva production via ultrasound-guided injection into parotid and/or submandibular glands [10, 11]. Though this minimally invasive technique is gaining popularity, the neurotoxic effects are temporary [10, 11].

Speech therapy is an alternative treatment option for sialorrhea, but there is no established timeline or treatment schedule for potential candidates. A recent literature review revealed a lack of studies on the outcomes of speech therapy on saliva control [12]. Training oral motor skills and improving sensory awareness in speech therapy has been shown to effectively reduce sialorrhea in patients with cerebral palsy, especially with adjuvant procedures or anticholinergics [13].

Sialorrhea can have an immense impact on patient and caregiver QoL. Successful management of pediatric sialorrhea includes an improvement to DIS scores. To date, there is limited literature on the ability for speech therapy to improve QoL in the pediatric population. The objective of this study is to evaluate the effects of targeted speech therapy on QoL for caretakers of children with sialorrhea as described by DIS. We hypothesized that caregivers would attribute an improved QoL after the implementation of speech therapy sessions for sialorrhea. Further, a more intensive program would yield even stronger improvements in QoL outcomes.

2 | Methods

2.1 | Institutional Review Board Approval

This study was submitted to and approved by the University of Texas Southwestern (UTSW) Institutional Review Board (Study #42274).

2.2 | Data Collection

This is a single-institution retrospective case series that included patients from the secretion management clinic at a pediatric tertiary care facility. All patients who had completed one of the two available outpatient speech therapy programs were included. We also included patients for whom therapy was recommended but the caregivers opted for observation. Speech therapy programs were classified as intensive (4 sessions per week for 3 weeks) or less intensive (2 sessions per week for 3 months). These protocols were developed based on previous literature validating the DIS in children undergoing speech therapy [6]. Caregivers were presented the two protocol options and had the option to choose the one which best fit their schedule, encouraging compliance.

Regardless of the therapy schedule, similar therapeutic modalities were used. The two most common therapeutic modalities utilized were neuromuscular electrical stimulation (NMES) and facial taping. NMES involves the use of electrical stimulation to activate muscles through stimulation of intact peripheral motor nerves. Major treatment goals of this therapy are to strengthen weakened muscles, aiding in the recovery of oropharyngeal motor control [14]. Electrical stimulation is known to be more effective when performed in combination with exercise therapy, such as swallowing [14]. A 2012 case series by Rice, et al. suggests that NMES is an effective intervention for children with pharyngeal phase dysphagia [15]. Frequency and duration is child-specific and based on clinical judgment by the trained speech language pathologist (SLP). The initial assessment was done by the same SLP and a group of multiple SLPs with the similar training for pediatric patients with sialorrhea conducted therapy sessions.

Caregivers completed DIS questionnaires before the patients' initial treatment session and at their discharge session. The DIS is a 10-item questionnaire with the ability for caregivers to rate the extent to which their child's drooling impacts their lives and can be used as a proxy to describe QoL [6]. For each question, caregiver level of agreement is assigned a score between 1 (representing the least impact) and 10 (representing the most impact). Final DIS scores range from 10 to 100 with an increased score indicating that patients and caregivers are more severely impacted by sialorrhea (Table S1).

We then reviewed patient charts on the electronic medical record to gather demographic information such as age, race, and sex. Collected comorbidities included: cerebral palsy, hypoxic-ischemic encephalopathy (HIE), developmental delay, seizure disorders, cardiac disease, chronic lung disease, neuromuscular disease, and obstructive sleep apnea. Additionally, we determined if the patients had any other interventions during the study period, such as salivary BT-A injections or addition of pharmacologic agents. We excluded patients who did not complete the planned therapy regimen or the DIS surveys.

Study data were collected and managed using REDCap (Research Electronic Data Capture) electronic data capture tools hosted at UTSW Medical Center [16, 17].

2.3 | Statistical Analysis

Statistical analysis was performed to compare DIS scores before and after therapy with the software Stata (StataCorp. 2023. *Stata Statistical Software: Release 18*. College Station, TX: StataCorp LLC). Patients were separated into 3 groups: intensive therapy, less intensive therapy, and no therapy. We compared demographics, comorbidities, and pre- and post-treatment DIS scores between these groups.

Four patients did also have salivary gland BT-A injections during the study period, 3 in the control group and 1 in the intensive therapy group. Due to the small number of patients in this group, we did not pursue further subgroup analysis.

Eighteen patients had some pharmacologic intervention during the study period, 9 in the control group, 4 in the less intensive

therapy group, and 5 in the intensive therapy group. Again, due to the small number of patients in this group we did not pursue further subgroup analysis.

There were seven patients with a tracheostomy and ventilator dependence. Six of these patients were in the more intensive therapy cohort and 1 patient was in the cohort which did not receive therapy. Due to the small number, subgroup analysis was not performed. Future studies on this population may be useful as they may represent more severe symptoms.

Continuous variables were shown as means with standard deviations. Categorical variables were shown as counts with percentages along with Fisher's exact testing for significance. A student's t test and analysis of variance (ANOVA) were used to compare continuous variables. Statistical significance was set at $p < 0.05$. This study adhered to Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines for observational studies [18].

3 | Results

A total of 49 children met inclusion with an initial management approach of more intensive therapy for 33% ($n = 16$), less intensive therapy for 29% ($n = 14$), and no therapy for 39% ($n = 19$). Age at initial management was 6.5 years (SD: 3.7), 7.9 years (SD:

5.6), and 8.1 years (SD: 4.6) respectively among groups ($p = 0.57$). Table 1 describes baseline characteristics, which were similar across all three management approaches.

Table 2 compares DIS data by initial therapeutic groups. The mean baseline DIS was 63.5 (SD: 12.4) for more intensive, 51.9 (SD: 18.6) for less intensive, and 55.6 (SD: 18.8) for no therapy ($p = 0.17$). After a mean of 2.6 months (SD: 2.9), post-therapy DIS scores were 47.2 (SD: 18.0) for more intensive ($p = 0.006$), 39.1 (SD: 16.6) for less intensive ($p = 0.07$), and 54.5 (SD: 16.4) for no therapy ($p = 0.86$). Post-therapy DIS scores were different between the three treatment groups ($p = 0.04$), with more intensive therapy lower than no therapy ($p = 0.006$) but with no difference to less intensive therapy ($p = 0.99$). DIS were similar after less intensive and no therapy ($p = 0.06$). Further, the difference in DIS scores was -16.3 (SD: 14.3) after more intensive therapy, -12.8 (SD: 14.3) after less intensive therapy, and -1.1 (SD: 12.6) after no therapy ($p = 0.005$). This is also portrayed in Figure 1.

4 | Discussion

This study is the first to explore the optimal speech therapy frequency on the QoL of patients with sialorrhea and their caregivers as described by DIS. A 2019 study by Reid, et al. validated the DIS via assessment of patient improvement in speech therapy, however no specific speech therapy timeline or regimen was recommended

TABLE 1 | Baseline characteristics by initial sialorrhea therapeutic approach.

Characteristic	More intensive therapy	Less intensive therapy	No therapy	P
Total, n (%)	16 (33)	14 (29)	19 (39)	—
Males, n (%)	12 (75)	9 (64)	8 (42)	0.14
Age, y. (SD)	6.5 (3.7)	7.9 (5.6)	8.1 (4.6)	0.57
Race, n (%)				
White	11 (69)	7 (50)	8 (42)	0.31
Black/African American	4 (25)	2 (14)	8 (42)	
Asian	0 (0)	2 (14)	1 (5)	
Other	1 (6)	3 (21)	2 (11)	
Hispanic ethnicity, n (%)	8 (50)	7 (50)	5 (26)	0.27
Short gestation, n (%)	4 (25)	4 (29)	6 (32)	0.93
Cerebral palsy, n (%)	7 (44)	7 (50)	13 (68)	0.35
HIE, n (%)	11 (69)	7 (50)	14 (74)	0.42
Developmental delay, n (%)	15 (94)	13 (93)	19 (100)	0.52
Seizure disorder, n (%)	11 (69)	7 (50)	17 (89)	0.05
Cardiac disease, n (%)	3 (19)	3 (21)	5 (26)	0.91
Chronic lung disease, n (%)	5 (31)	5 (36)	8 (42)	0.93
Aspiration, n (%)	9 (56)	7 (54)	11 (58)	0.99
Neuromuscular disorder, n (%)	10 (63)	6 (43)	11 (58)	0.59
Obstructive sleep apnea, n (%)	12 (75)	7 (50)	8 (42)	0.15

Abbreviation: SD, standard deviation, Hypoxic-ischemic encephalopathy.

TABLE 2 | Drooling impact scales by initial sialorrhea therapeutic approach.

Variable	More intensive therapy (n = 16)	Less intensive therapy (n = 14)	No therapy (n = 19)	P
Baseline DIS, mean (SD)	63.5 (12.4)	51.9 (18.6)	55.6 (18.8)	0.17
Therapy time, m (SD)	2.6 (2.4)	2.9 (2.6)	2.3 (3.4)	0.84
Post-Therapy DIS, mean (SD)	47.2 (18.0)	39.1 (16.6)	54.5 (16.4)	0.04
DIS difference, mean (SD)	−16.3 (14.3)	−12.8 (14.3)	−1.1 (12.6)	0.005

Abbreviations: DIS, Drooling Impact Scale; SD, standard deviation.

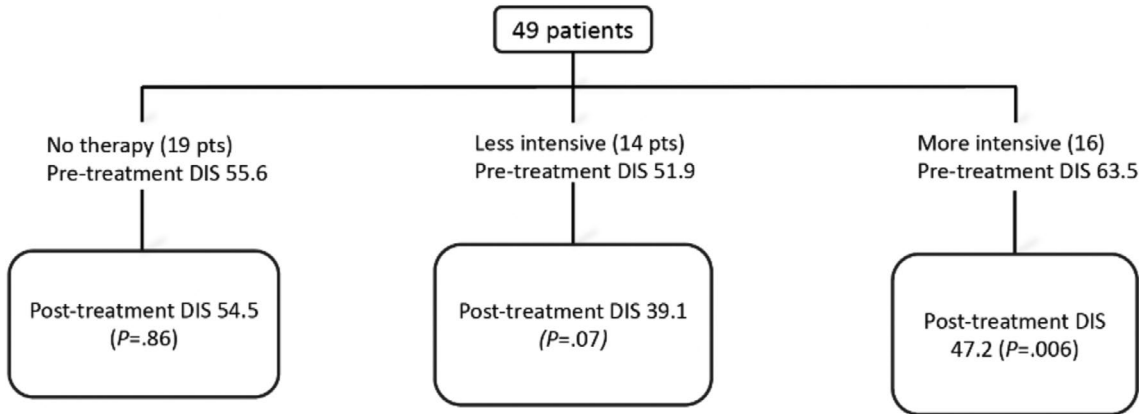


FIGURE 1 | Flow chart showing treatment groups and pre-and post-treatment Drooling Impact Scale (DIS) scores.

[6]. Our findings are in line with this study, finding that participation in speech therapy significantly improved DIS [6]. This may be interpreted as a proxy for QoL, describing improved drooling impact and QoL for children with sialorrhea and their caregivers. Importantly, the present study determined that there was more improvement in patients who underwent the intensive therapy regimen compared to the less intensive regimen.

4.1 | Impact of Sialorrhea on Quality of Life

Sialorrhea greatly impacts the QoL of patients and caregivers alike [4, 5]. DIS is an important measure as it addresses well-being of sialorrhea patients along with those most involved in their care [6]. As demonstrated in this study, patients experiencing sialorrhea are often medically complex with significant neurological comorbidities. Management of sialorrhea is vital for patient safety as well, due to the increased risk of aspiration pneumonia and respiratory complications associated with poor secretion management [19].

4.2 | Utility of Speech Therapy in Sialorrhea

The benefits of speech therapy on swallowing dysfunction have been well-studied, however the impact on drooling and secretions is minimal [12]. Previous studies have supported the use of behavioral interventions, such as token economy reinforcement, positive reinforcement, prompting, self-control, and over-correction [20]. Though improved sialorrhea was reported, there was no standardized outcome measure between

studies and impact on patient and caregivers using DIS was not reported.

The results of the present study demonstrate a clear benefit of speech therapy for sialorrhea patients. Though the intensive regimen led to a greater decrease in DIS when compared to the less intensive, both options had significantly greater impacts than no speech therapy. The availability of two effective timelines may improve patient compliance with speech therapy due to the ability to accommodate caregiver schedules [21].

Reid et al. demonstrated that speech therapy yielded less improvement in DIS compared to salivary BT-A injections [22]. However, efficacy of BT-A in sialorrhea management decreases over time while the goal for speech therapy is longitudinal improvement and management [22]. Additionally, a study comparing salivary BT-A injections with scopolamine medical management identified similar success rates between the two modalities utilizing scales other than DIS [23]. These results highlight the importance of multimodal sialorrhea management. We recommend that speech therapy should be considered as a conservative first line approach to managing sialorrhea. This is due to its efficacy and minimal risk of adverse effects when compared to medical or surgical interventions [8, 9].

Various interventions may be utilized in speech therapy, such as NMES and facial taping. NMES stimulates contraction of the muscles involved in swallowing, allowing for improved muscle strength, control, and secretion management [14, 24]. Though research on the efficacy of NMES in treating dysphagia presents varying conclusions, it has demonstrated potential in the

management of sialorrhea in smaller studies [24, 25]. Facial taping is a speech therapy modality which has demonstrated a significant reduction in sialorrhea among pediatric patients with intellectual disability [26].

4.3 | Strengths and Limitations

There are several strengths to this study. It is the first study to thoroughly analyze the impact of structured speech therapy on patients with sialorrhea [6, 7]. It also identified the most effective therapeutic schedules by comparing an intensive and less intensive option to no therapy. Importantly, there are limitations to this study. The uncontrolled, open-label methodology in this retrospective analysis poses risk for bias and confounders. Therefore, this data would be further supported by a prospective controlled study of the impact of speech therapy on DIS. This was also a small patient cohort. There are several confounders such as pharmacologic intervention, salivary gland botulism injections, and severity of comorbidities that we were unable to analyze due to small numbers. Additionally, sialorrhea is a possible side effect from seizure medications and the extent of patients' seizure disorders or neurological disease lack characterization in this study. Caregiver self-selection may also be a source of selection bias and emphasizes the need for future randomized controlled trials on this topic. Future research may include more variables relating to sialorrhea and patient functional status, to create a more well-rounded understanding of how this condition is impacted by speech therapy. Additionally, future research may describe the sustainability of improvement following completion of speech therapy as well as best maintenance practices.

5 | Conclusion

Speech therapy is a conservative, non-invasive treatment for sialorrhea, improving patient and caregiver quality of life as described by DIS. An intensive program of 4 sessions per week for 3 weeks is more effective than a less intensive regimen of 2 sessions per week for 3 months. However, both programs yield improved results compared to patients who did not complete speech therapy. Future research may focus on optimal speech therapy techniques in sialorrhea management.

Conflicts of Interest

Dr. Romaine Fitzgerald Johnson is the editor-in-chief of *Laryngoscope Investigative Otolaryngology*. The other authors declare no conflicts of interest.

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Supporting Information

Additional supporting information can be found online in the Supporting Information section.