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Efficacy of Oral-Sensory-Motor Intervention in The Management for Drooling of Saliva in Children with Cerebral Palsy - A Cross-Sectional Study

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ABSTRACT

Purpose: Drooling of Saliva is one of the major issues that can have a serious effect on the health of those children suffering from it which is often prevalent in children with CP but seldom looked into during therapeutic intervention. Hence, we focused on treating the drooling through oral-sensory-motor intervention (oral-motor therapy + rMV) instead of a medical/surgical intervention and to also assess its efficacy as a management option.
Methodology: This is an observational study, administered on 13 children of 5 to 16 years, diagnosed with severe drooling consequent to CP. The efficacy was assessed by comparing the baseline & follow-up scores measured using the Drooling Impact Scale (Reid et al., 2010).
Result: The results showed that there was a statistically significant improvement ('t' critical=2.179; p-value ≤ 0.05) in all the 10 parameters. No statistically significant difference was observed with respect to age or type of CP.
Conclusion: The study concludes that oral-sensory-motor integration proves to be an effective management to control the drooling of saliva in children with CP. However, to understand the influence of the factors like age or type of CP, further research with larger sample size is warranted.

Keywords: Cerebral Palsy; Drooling; Behavioural Integration; Sensory-motor Integration; Drooling Impact Scale; Oro-motor exercises

INTRODUCTION

Saliva is a watery fluid that is secreted by the salivary glands in the oral cavity. Saliva is very important as it greases the bolus of food in our mouth, mixes it and helps in swallowing the food with ease. Also, saliva helps partly with the breaking down of the nutrients in the food bolus while chewing. The condition in which excessive pooling of the saliva in the antero-inferior oral cavity which may drool out causing a distress is termed as drooling of saliva. This condition occurs when the oral-facial and palate-lingual muscular structures lose the ability to control the secretions through co-ordinated movements and not because of superfluous saliva secretions (Meningaud et al., 2006). Drooling along with causing physical and cosmetic discomfort, affects the control over the food, inside the oral cavity during chewing and swallowing and this in turn may lead to aspiration pneumonia and/or malnourishment due to inadequate food intake, affects speech production including the disturbance in the articulation and in the flow (pauses might occur during the speech due to drooling, disrupting the continuity) of speech (Meningaud et al., 2006; Speyer et al., 2019).

Cerebral Palsy (CP) is one of the most common motoric disorder in children that affects their posture and makes it difficult for them to balance and/or make a movement (Centers for Disease Control and Prevention, 2021). The overall pooled prevalence of CP of those who were born after 1985 was found to be 2.11 per 1000 births through a meta-analysis of 13 studies (Oskoui et al., 2013) There are different types of Cerebral Palsy like Spastic, Hypotonic, Ataxic, Athetoid and mixed types. These children also suffer from drooling disorder (Speyer et al., 2019; Centers for Disease Control and Prevention, 2021). A prevalence study by Tahmassebi and Curzon in 2003, showed that 58% (93 of 160 children with CP) had drooling of saliva with 33% of children having severe drooling (Tahmassebi & Curzon, 2003). A meta-analysis of 42 studies by Speyer et. al., 2019 showed a 44% pooled prevalence (high prevalence) of drooling of saliva in children with CP worldwide (Speyer et al., 2019). From the evidences revealed by the literature, it becomes obvious that drooling is indeed a very vital component especially in children with CP that has to be treated along with other primary interventions that are carried usually carried out.

The usual procedures to intervention are by injecting BoNT-A into the salivary glands or through anticholinergic medications (Collins et al., 2020; Reid et al., 2010). However, in many clinical and rehabilitation set-ups, drooling has been treated through behavioural interventions and the preference of the caretakers or parents, towards such interventions are increasing. But there is dearth of evidences for effectiveness based on the type of intervention and the structured intervention practices are comparatively lower, for the cerebral palsied population in India. Also, repeated muscle vibration (rMV) technique is adopted into the new trends of treating CP related drooling issues. The impulses generated by rMV induces both a somatosensory and a motor reaction by stimulating the corresponding central structures which helps in achieving control over the oral-motor structures as well in obtaining improvements in swallowing functions (Russo et al., 2019).

Therefore, in this study, we aim to analyse the effectiveness of the rMV when combined with the traditional oral-motor therapy for drooling in CP children. Also, this study records the severity of drooling and its impact on children with different types of CP and their families.

Aim

The primary aim of the study is to understand if the oral-sensory-motor therapy is effective in controlling the drooling of saliva in children with different types of Cerebral Palsy.

Objectives

1. To check and quantify the effectiveness of oral-sensory-motor therapy as an intervention to reduce drooling of saliva in children with CP.
2. To compare the effect of drooling intervention between the children of different Cerebral Palsy types.
3. To compare the effect of drooling intervention between the different age groups of the participants in the study population.

METHODOLOGY

Study Design

This is a cross-sectional study to understand the effect of intervention on drooling of Saliva in a small district of North Western India.

Study setting

The study was conducted at the department of Audiology and Speech Language Pathology, Smt. P.N.R. Society for Relief and Rehabilitation of the Disabled (NGO), in Bhavnagar, Gujarat, on the 31st of July, 2021. The study was approved by the organisation ethical committee.

Participants

13 Children with drooling disorder who were diagnosed to have Cerebral Palsy with no co-morbid conditions were taken as the

study participants. The participants aged between of 5 to 16 years (mean age = 9.15 years) of both male and female gender groups. The participants were grouped based on their age groups and the type of Cerebral Palsy as shown in Table 1. The children with associated conditions such as visual or hearing impairment and other associated conditions such as ASD, ADHD, etc. were excluded from the study.

Age group	Type of CP			Total
	Spastic	Flaccid	Mixed	
4-7 years	2	1	1	4
7-10 years	0	1	2	3
10-13 years	2	0	1	3
13-16 years	2	1	0	3
Total	6	3	4	13

None of the children underwent any previous intervention for drooling and were monitored by special educators and SLPs throughout the intervention period. All the participants were involved in the study only after receiving a signed consent from their parents/caretakers to whom the purpose of the study and the procedures were detailed.

MATERIALS USED

For Assessment

As per the biological-psychological-social model of disability postulated by ICF-10, in 2001 (Rauch et al., 2008; Wenzel & Morfeld, 2016), in order to provide a holistic improvement through an appropriate intervention method, information on body structures and functions (primarily on oral, in this study), general and oral health related to personal and environmental factors, was obtained from each participants through a detailed case history, Oro-motor checklist and Drooling Impact Scale (DIS), a rating scale to understand the severity of drooling and its impact, was used.

Case history. A complete case history with information starting from demographic data to the provisional diagnosis, including birth history, medical history, social and behavioural history was procured through a direct interview with the parent/caretaker.

Oro-motor checklist. We also used the reflex and oral-motor checklist to serve as the baseline to plan the intervention for each participant. It profiles the structural and functional abilities of oral structures such as lips, tongue and jaw on the basis of position at rest as 'open' or 'clenched' or 'protruded' or 'appropriate', strength as 'weak' or 'strong', range of motion (for jaw and tongue) as 'restricted' or 'appropriate' and tonicity (for tongue and lips); basic reflexes such as tongue reflex, biting reflex, and gag reflex as 'presence' or 'absence', drooling based on the severity; vegetative functions such as blowing, sucking, chewing and sucking; other parameters such as obtention of neck control, food aversion, respiratory support and ambulation.

Drooling Impact Scale. For the baseline assessment and to understand if the intervention was effective after the stipulated period of time, the DIS was administered (Reid et al., 2010). It consists of 10 questions that taps on the frequency of the child's drooling and its impact on the child and family, on a 10-point rating scale. The DIS has good test- retest reliability. It was found to have a good correlation (>0.5) with the carer's global rating of change in drooling and good validity. Also, this is useful for testing the effectiveness of intervention as it is sensitive even for a change within a short period of intervention and the reduction in scores can be utilized as a prognostic indicator (Reid et al., 2010).

The reduction in scores from the baseline on the DIS after intervention, were interpreted to comment on the effectiveness of the treatment as per the guidelines given by Reid et al., in 2010 (Reid et al., 2010).

For Intervention

We used the sensorimotor integration strategies and Oro-motor exercises such as range of motion exercises, resistance exercises and falsetto exercises for treating the drooling of saliva (Arvedson, 1998; Mcmurtrey, 2007; Miller & Willging, 2013). For sensory stimulation during oral-motor integration, we used the Z-vibe® tool, manufactured and marketed by ARK Therapeutics, USA (ARK Therapeutic -

Makers of Innovative Special Needs Products & Sensory Therapy Tools).

PROCEDURE

All the participants were administered with a detailed case history and the individual's Oro-motor structure and function and reflex behaviours were profiled in order to understand the individual's defect/difficulty and to plan the intervention accordingly before initiating the course of intervention.

A baseline drooling assessment was conducted using the Drooling Impact Scale. The intervention was planned for a period of 1 month (Reid et al., 2010), accounting to a total of eight, 30 minutes sessions, pre-scheduled for each individual participant with adequate inter-session intervals. The intervention began with the manual oral-motor exercises to improve the strength of the oral muscles as well as to facilitate adequate motion. The exercises included isometric and isotonic exercises for the lip, tongue and the jaw (including, tongue extension, retraction and lateralization with and without resistance, puffing up of check by inducing an intra-oral pressure, oral-facial massages) These exercises also help in improving lingua-palatal strength that helps in bolus formation and swallowing. Each exercise was repeated for 10 times. Followed by the manual oral-motor exercises, sensory stimulation of the labial, lingual, mandibulofacial and maxillofacial muscles were provided through Z-vibe® tool, for 15 minutes, to facilitate the lip closure, tonal improvement, posture, sensory abilities, speech and swallowing.

All the participants were monitored during the entire course of intervention by the special educators and SLPs. The prognosis was checked on a regular basis during the course of intervention and the final assessment was done similar to the baseline after the completion of 8-session intervention.

Statistical Analysis

In order to compare the improvement of the symptoms (reduction in drooling and its consequences), paired 't' test was performed on the pre- and post- intervention scores assessed using the drooling impact scale. Paired 't' test was performed for each question as well as on the total scores to check if the improvement observed were statistically significant. Difference in the group mean scores were also calculated and compared between the 3 groups of participants who were divided based on the type of CP to understand the relationship between the type of CP and the prognosis. To compare the prognosis between the groups, statistically, one-way ANOVA was used.

RESULTS

Baseline Assessment

A detailed case history was administered on all the participants. Then, a basic oral-reflex profile and Oro-motor profile was recorded for each participant using an Oro-motor checklist. The data showed that almost all the participants had Oro-pharyngeal dysphagia and the strength of the oral structures such as lips, tongue and jaw were weak. The range of motion was also restricted in most of the participants. More than 75% of the participants had poor vegetative functions such as blowing (92.3%), sucking (61.54%) and chewing (84.6%). More than half (61.54%) the participants had inadequate respiratory support.

The DIS was administered as a part the baseline assessment to understand the severity and the impact of drooling on the child and his/her family. The scores were recorded for each question on a total of 10 and their mean scores are shown in Figure 1 and an overall score on a total possible score of 100{[Total no. of questions (i.e,10)] * [max obtainable points (i.e,10) = 100)] as in Figure 2.

Figure 1: The group average scores obtained on DIS during the baseline assessment for each question is represented.

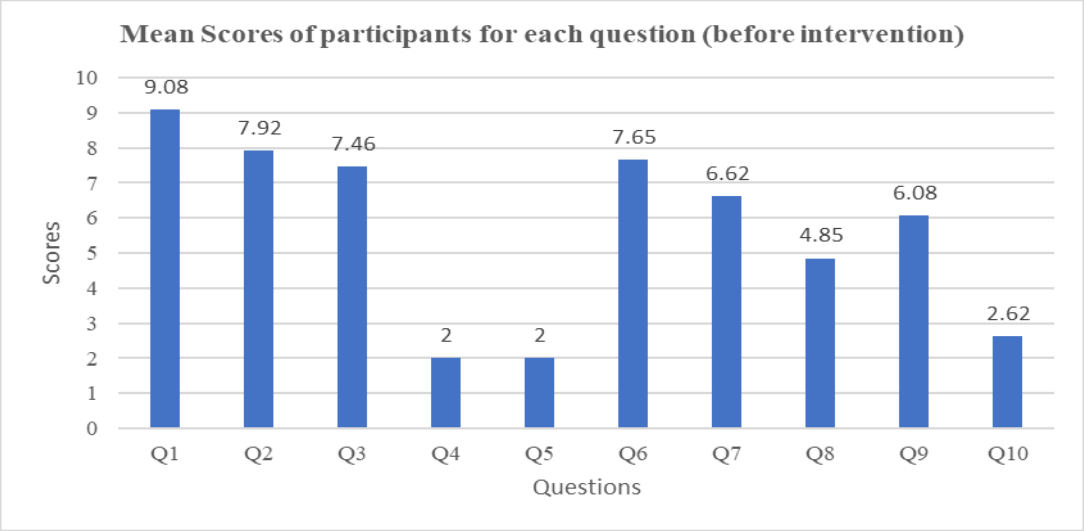
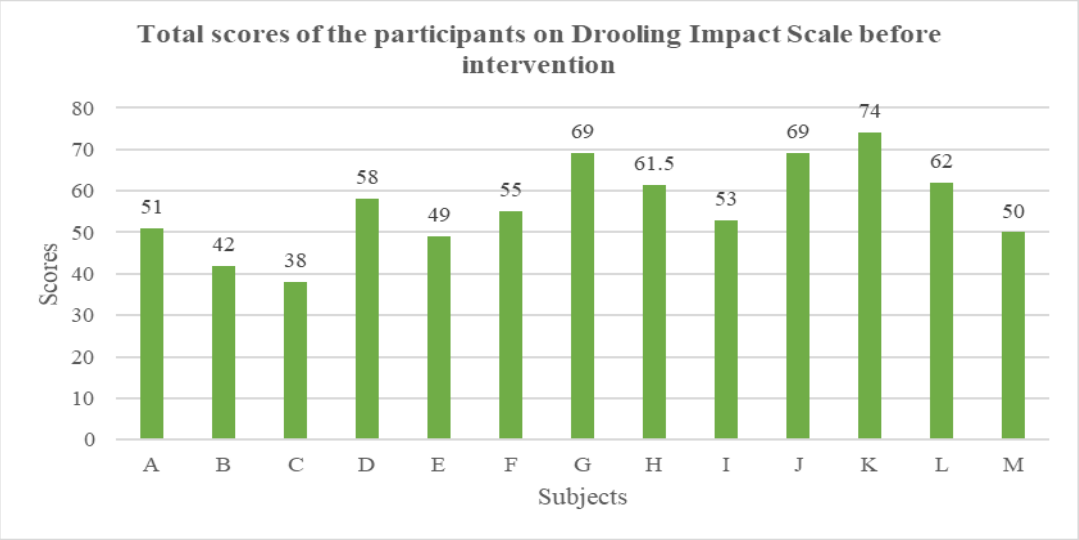


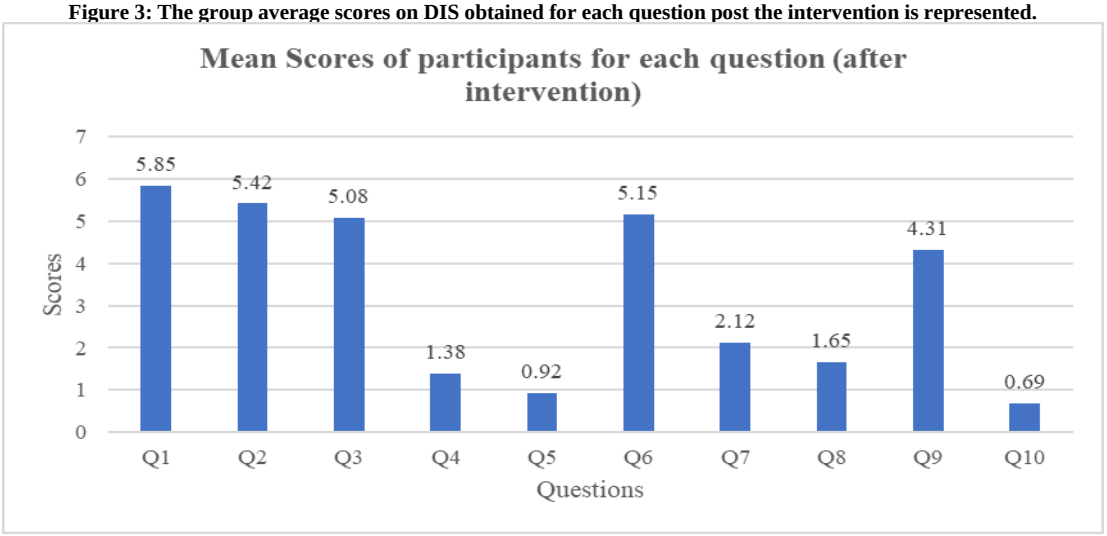
Figure 2: The total scores obtained by each individual on DIS before intervention is represented



Post-Intervention

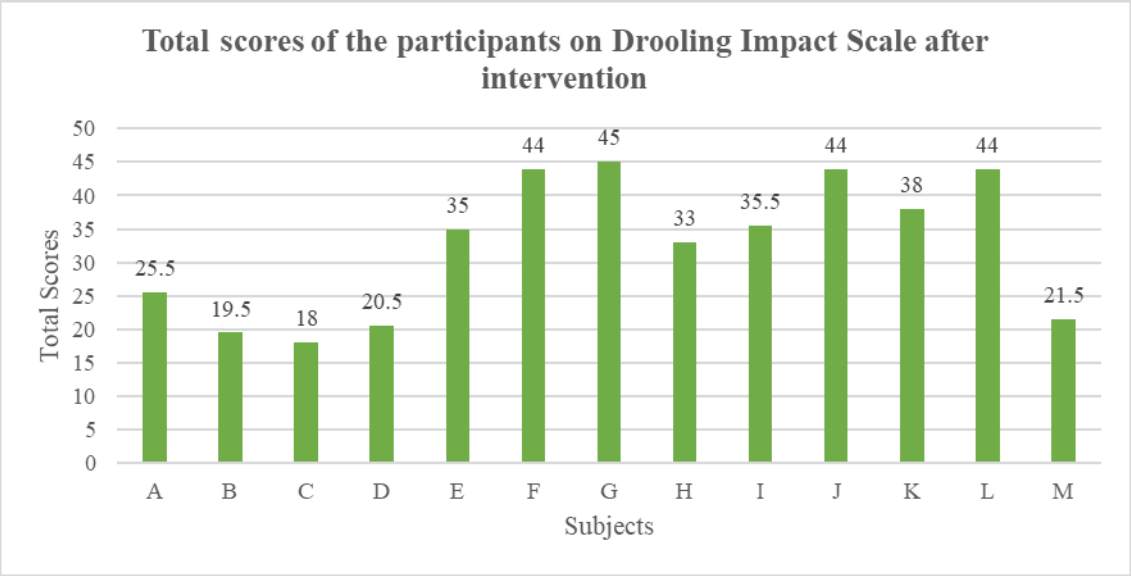
After a period of 1 month, a follow-up evaluation was done for all the participants in the study. The mean scores obtained for each question on the DIS from the participants are shown in Figure 3.

Figure 3: The group average scores on DIS obtained for each question post the intervention is represented.



The total scores of each participant were also calculated on a total score of 100 similar to the baseline evaluation as represented in Figure 4.

Figure 4: The total scores obtained by each individual on DIS post intervention is represented



Effectiveness of Intervention

To understand how much the intervention has contributed to the betterment/improvement in controlling drooling and reducing its impact, a paired comparison of pre and post intervention scores for each question and overall scares were calculated, which showed a very good improvement. Paired 't' test was used to check for statistical significance of this measure and it showed the reduction in scores for each question and the total scores were statistically significant ($p \leq 0.05$). More than 80% of the pairs even showed that the reduction in scores were highly statistically significant (p value ≤ 0.001) which clearly shows that the therapeutic strategies were useful for the participants in reducing the drooling of saliva (Table 2).

Table 2 shows the statistical comparison between the pre and post intervention scores obtained on Drooling Impact Scale

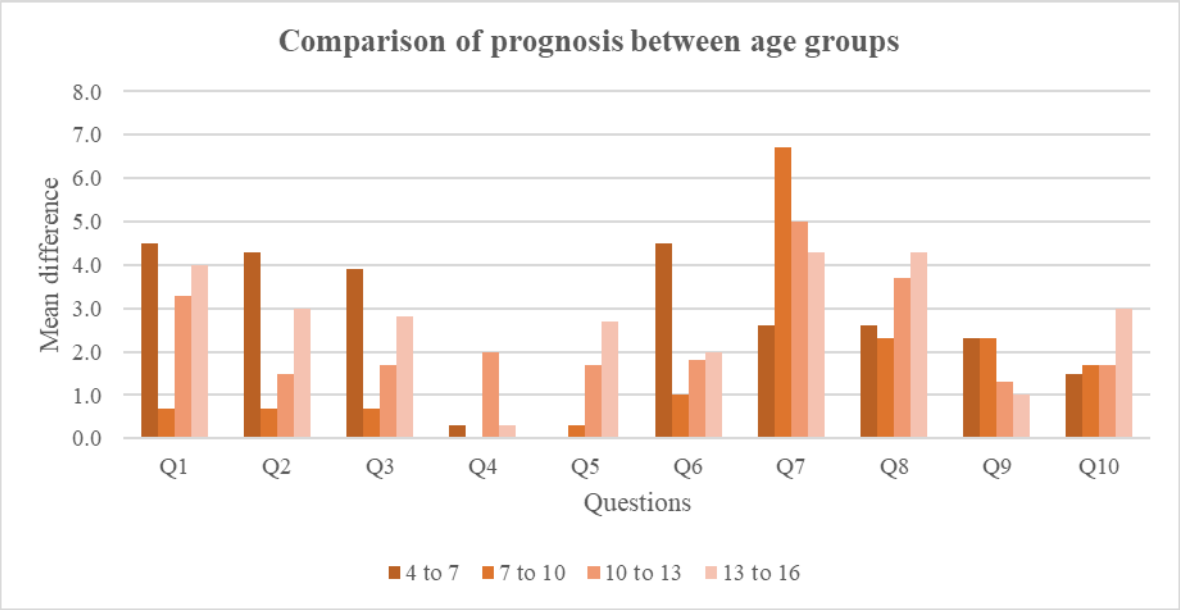
Paired 't' test on Drooling Impact Scale							
Pair	**Sample/Item**	**Before/After**	**Frequency**	**Mean**	**Std. Deviation**	**t-value**	**p-value**
1	Q1	Before	13	9.08	1.07	6.697	0.000**
		After	13	5.85	2.03		
2	Q2	Before	13	7.92	1.07	5.395	0.000**
		After	13	5.42	1.63		
3	Q3	Before	13	7.46	1.01	5.300	0.000**
		After	13	5.08	1.74		
4	Q4	Before	13	2.00	2.08	2.309	0.040*
		After	13	1.38	1.60		
5	Q5	Before	13	2.00	2.72	2.419	0.032*
		After	13	0.92	1.21		
6	Q6	Before	13	7.65	1.59	5.204	0.000**
		After	13	5.15	2.16		
7	Q7	Before	13	6.62	1.39	6.624	0.000**
		After	13	2.12	1.42		
8	Q8	Before	13	4.85	2.11	5.489	0.000**
		After	13	1.65	1.03		
9	Q9	Before	13	6.08	3.41	4.153	0.001**
		After	13	4.31	2.61		
10	Q10	Before	13	2.62	2.53	3.755	0.003*
		After	13	0.69	0.99		
11	Total Scores	Before	13	56.27	10.27	10.468	0.000**
		After	13	32.58	9.97		

Note: 't' critical=2.179
***Statistically significant (p value ≤ 0.05); **highly statistically significant (p value ≤ 0.001)**

Factors affecting the intervention

Age

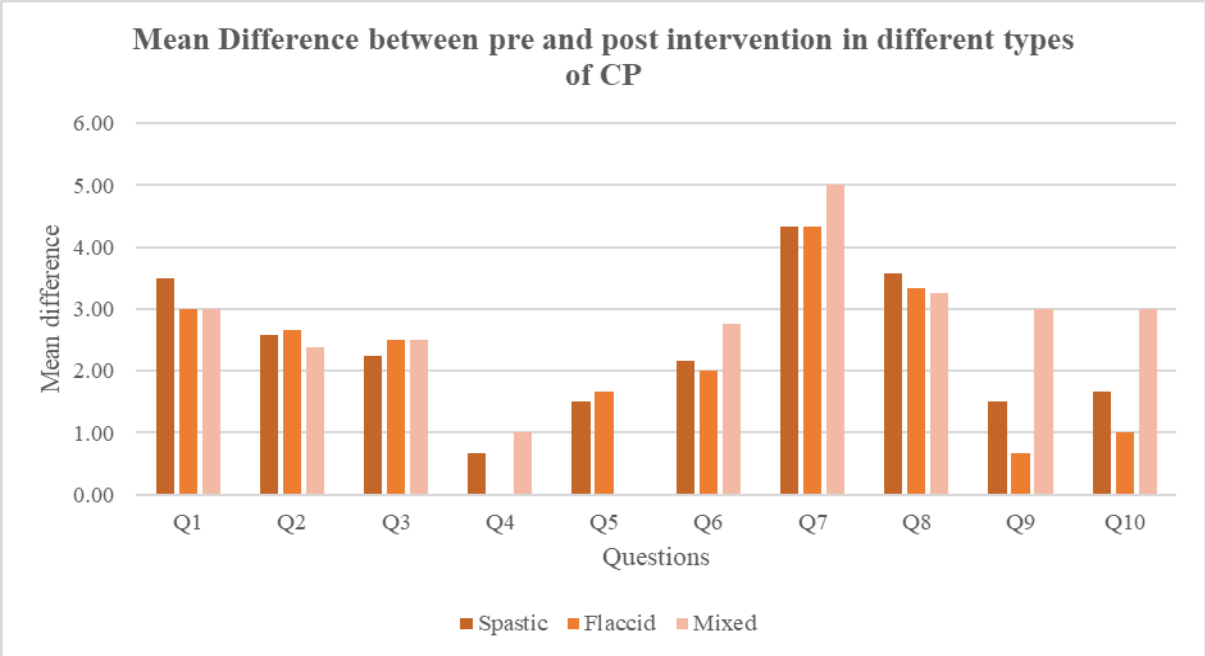
Figure 5 represents the difference in improvement between the 4 different age groups.



To analyse if there is an effect of age, the difference in the mean scores of each group were compared (as seen in Figure 5). It shows that there is a difference between the age groups in the difference between the pre and post intervention scores. When compared the overall difference between the pre and post intervention scores, its observed that the eldest group in the study population (aged 13 to 16 years) showed the maximum overall improvement, followed by the youngest group (aged 4 to 7 years). However, this observation was not statistically significant when analysed using one-way ANOVA (p value ≤ 0.05).

Type of CP

Figure 6 represents the difference in improvement between the groups of spastics, flaccid and mixed types of CP.



To understand if there is an effect of the type of CP in the prognostic improvements, the difference in their individual scores were calculated and averaged across the three groups. The results revealed that there is a slight difference between the groups in each aspect on the scores of Drooling Impact Scale. The improvements are somewhat similar for Spastic and Flaccid types as inferred from the Figure 6. However, this was not proved to be statistically significant.

DISCUSSION

Drooling of Saliva is one of the major issues in children, especially in those children with CP. But it is not really treated with primary importance. Drooling is stated to be in relation to or a consequence of swallowing difficulties rather than hypersalivation in children with CP (Erasmus et al., 2009; Senner et al., 2004). In our study, we primarily focussed on treating drooling of saliva and also worked to improve the swallowing abilities in children which may passively help control pooling of excessive saliva.

To control the drooling of saliva, we used rMV approach which has been observed to show good clinical improvements in the recent years, even for feeding difficulties, along with oral-motor exercises (Russo et al., 2019; Sergio machado et al., 2010). This is due to the assumption that improving the sensory experience of the child may yield better activation and control over the structures that contribute towards the problem and hence, betterment. We used Z-vibe® tool from ARK Therapeutics, USA, to induce a sensory stimulation through tactile and vibration modality, which when combined with verbal instructions and apt reinforcements, yielded improvement on lip closure, muscle tone, strength and control and coordination. The sensory intervention also helped overcome feeding and swallowing issues to an extent, with regards to oral-preparatory and oral-transit phase.

To understand the effectiveness, we compared the scores of the baseline with post-intervention scores of Drooling Impact Scale, and it showed great improvement which was statistically significant (p value $\leq$ 0.05). It is also important to note that few aspects like the smell of the drool, may not really change with the therapy, yet, it may reduce in its pungency, due to the reduction of the severity. The effect size, calculated based on the statistical criteria given by Jacob Cohen (Jacob Cohen, 2013; Reid et al., 2010), was also large for the participants in this study showing that there was very good improvement through the intervention for the participants. This is in agreement with the previous study findings on rMV that there was a statistically significant improvement ($p<0.05$) in DIS, Drooling quotient (DQ), Visual Analog Scale (VAS) and Drooling Frequency and Severity Scale (DFSS) in comparison to the baseline measure when used an electromechanical transducer with mechanical support to apply pressure over the sub mandibular muscles for three consecutive days at regular intervals of time (Russo et al., 2019).

The main factors that affect the treatment outcome were age, type of CP, severity and oral motor control (Reid et al., 2012; Thomas-Stonell & Greenberg, 1988). In this study, we have compared two of these factors – age, types of CP. The mean difference in the scores of participants across age groups and types of CP were calculated. Although there was a change in the mean difference in scores, it was not statistically significant. But this may be due to the fact that the study population was limited, meaning, the sample size was small. Overall, the primary aim of the study was fulfilled revealing that the oral-sensory-motor therapy was very useful in treating the drooling of saliva in children with different types of CP.

CONCLUSION

The study concludes that oral-sensory-motor therapeutic approach does prove to be useful for controlling/managing the drooling of saliva and also the related feeding issues, when the intervention is appropriately planned for each individual. Oral-sensory-

motor therapies can be provided in rehabilitation centres and special schools and can serve to be a reliable and effective option for the management of drooling disorders in children who do not wish to opt for surgical/medical interventions. However, subsequent studies with large sample size are warranted, in order to truly understand the factors contributing to the treatment outcome like age, type of CP, etc.

Declaration of Interest: The authors report there are no competing interests to declare.

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Ethical Approval: Approved

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