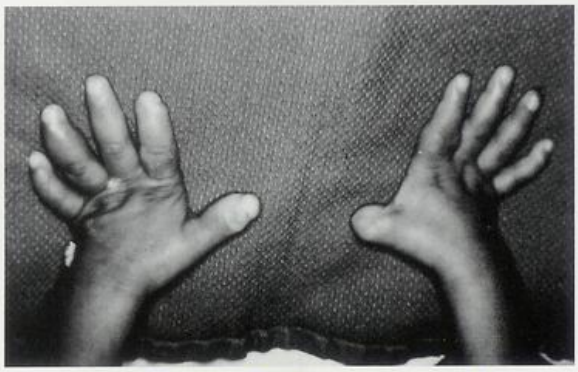
PEDIATRIC NEUROLOGY



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Traditional Chinese Medicine (Tongue Acupuncture) in Children With Drooling Problems

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Tongue acupuncture is an innovative technique in traditional Chinese medicine. We have demonstrated that specific tongue acupoints are related to various functional domains. This study aimed to assess the efficacy of tongue acupuncture in children with neurologic disability who had severe drooling problems. We conducted an intent-to-treat study in a cohort of 10 children. A continuous course of tongue acupuncture was performed daily to five acupoints in the tongue for a total of 30 sessions. Standardized outcome measures of drooling were evaluated by a blinded assessor to study the efficacy at baseline and after a course of treatment. Statistically significant improvement was noted in the following outcome measures: (1) mean visual analog scale (VAS) decreased from 6.6 (pre-TAC) to 4.67 (post-TAC) ($P = 0.002$); (2) mean drooling quotient (DQ) decreased from 14.016% (pre-TAC) to 8.335% (post-TAC) ($P = 0.0078$); and (3) mean drooling score (DS) decreased from 7.4 (pre-TAC) to 4.4 (post-TAC) ($P = 0.002$). This study demonstrated the efficacy of tongue acupuncture as an adjunctive or alternative treatment for patients with drooling problems and can be integrated as part of the oromotor stimulation program, drooling program, and behavioral modification program before subjecting the patient to invasive surgical procedures on the salivary glands.

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Introduction

Drooling (siglorrhea) is normal in young infants; it usually subsides by 18 months a as result of physiologic maturity of the oromotor function [1]. Drooling beyond 4 years of age is considered abnormal. Persistent drooling is socially stigmatizing and may affect interpersonal relationship because of bad odors and unhygienic appearance. Persistent drooling may be due to infrequent swallowing, inadequate lip closure, or poor head posture [2]. Drooling primarily results from an overflow of saliva from the mouth because of dysfunctional voluntary oral motor activity, improper swallowing, or oral sphincter deficits, but rarely because of hypersalivation.

Children with neurologic disorders, such as cerebral palsy, neuromuscular disease, autism, language disorders, or mental retardation have drooling problems. It is estimated that 10% of children with cerebral palsy have drooling problems [1,3-6].

For a drooling-management program to be successful, a multidisciplinary drooling team is essential for assessment and management of drooling [7-11]. Many methods, singly or in combination, have been used for treating the problem of drooling. However, none have been successful universally. Methods for treating drooling problems include the following: (1) oral motor stimulation program, which involves the stimulation of oral motor coordination and proprioception; (2) behavioral modification program, which uses a conditioning technique with auditory or visual signals as cues to prompt swallowing [12-14]; (3) medication, such as anticholinergic drugs [15,16]; and (4) surgical removal of salivary glands or salivary duct relo-

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cation for patients with severe oromotor dysfunction or cognitive deficits [17].

Oral motor and/or behavioral intervention depends on the intellectual ability and motivation of the child and is quite time consuming. Chronic use of anticholinergic drugs have side effects, such as constipation, bradycardia, reduced bronchial secretions, urinary urgency and retention, dilatation of pupils, loss of accommodation, photophobia, dry mouth, and flushing and dryness of skin [18]. Postoperative results of salivary duct ligation or relocation may lead to dryness of the mouth and an increase in the incidence of dental decay [19,20].

Quantitative measurement of drooling in children is difficult. Drooling frequency and severity varies daily and is influenced by hunger, hydration, emotional state, level of wakefulness, and other concurrent medication being given. Investigations into the mechanism of drooling are available [21-26].

In traditional Chinese acupuncture, which has been practiced in China for over two millennia, 400 acupoints on the body surface are interrelated to various functions. The surface acupoints are linked through 14 meridians to various organs and viscera of the human body. The approach in traditional Chinese medicine, in sharp contrast to western medical concept, is a "holistic" approach with a more philosophical background of balancing the "yin" and "yang." The physiologic basis of traditional Chinese medicine is aimed to improve "energy" or "body-flow" (so-called de-qui in Chinese). Even a normal human subject will respond to acupuncture resulting from the flow of energy. The effect of acupuncture was hypothesized and proven in animal and human studies to result from direct neural stimulation; changes in neurotransmitters, such as endorphin; immunologic markers; or endocrinologic signals. Thus acupuncture is especially effective in chronic disorders, especially in brain disorders [27].

Tongue acupuncture (TAC) is an innovative acupuncture technique invented by Dr. Sun, who practiced this technique in Peoples' Republic of China. After over 20 years of clinical experience, he discovered that there are at least 40 acupoints on the tongue, which are linked through 14 meridians to various organs and viscera, according to traditional Chinese medicine concept. Specific acupoints subserve various functional domains of the human body, similar to "the little man in the motor and sensory cortex." Clinical efficacy of acupuncturing the surface or base of the tongue in specific acupoints has proved the efficacy in improving various functional modalities for patients with chronic neurologic disorders. Our clinical experience with TAC, using objective outcome measures, has demonstrated functional improvement in pseudobulbar palsy and motor function in adults with stroke and children with cerebral palsy [28-34].

This study aimed to assess the efficacy of TAC as an adjunctive treatment in children with severe drooling problems who failed conservative treatment.

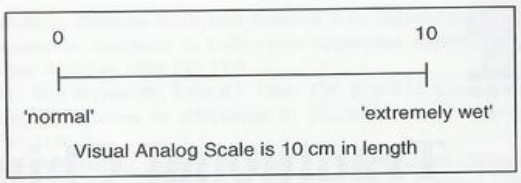

Figure 1. Visual analog scale.

Materials and Methods

A total of 10 children (five males and five females) with persistent drooling were recruited into the study as an intention-to-treat category. These children had been actively followed up in the multidisciplinary drooling clinic of the Children's Habilitation Institute of the Duchess of Kent Children's Hospital. Normal children, or those who drooled for unknown reasons other than physical or cognitive disability, were excluded. Children with bleeding disorders were excluded.

The children had severe physical and/or cognitive disability and were unable to comply with a formal behavioral modification program or oral-motor training program. They were unable to follow directions or imitate oral movements on command. The mothers were supportive in bringing the children back for daily treatment on an ambulatory basis for a consecutive period of 6 weeks.

Treatment

TAC was applied to five specific acupoints on the tongue daily by Dr. Sun using a sterile disposable 0.3 × 4-cm acupuncture needle (Made in China HWA TO). Two acupoints at the center of the tongue surface (TAC #1 = Run Ze' [1 cm from the tongue tip] and TAC #2 = Guan Zhu' [0.5 cm from tongue tip]) and three acupoints at the bottom of the tongue (i.e., sublingual region) (TAC #3 = Tian Men' [center of tongue base] and TAC#4 = Di You' [0.5 cm from tongue base on both sides]) were punctured. The tongue surface, TAC #1, was punctured by 1-cm depth obliquely, and TAC #2 was about 0.3-cm depth obliquely. The bottom of the tongue, TAC #3, was punctured perpendicularly by 0.5- to 1-cm depth, and TAC #4 was 0.5-cm depth perpendicularly.

The total acupuncturing procedure lasted for less than 15 seconds, and no sedation was required. The child sat on the mother's lap with the head tilted approximately 45 degrees upwards, and sterile gauze was used to pick up and station the tongue with the examiner's left hand. The child was encouraged to open up his/her mouth, which was the usual mouth posture of those with persistent drooling. Quick and accurate insertion into five acupoints was performed with the examiner's right hand. Most children tolerated the procedure well. A total of 30 sessions (daily sessions for a total course of 6 weeks was administered). The children attended our Tongue Acupuncture Research Clinic on an ambulatory basis and were discharged immediately after TAC.

Measurement of Drooling

Drooling was assessed by the following three techniques, which have been validated in other studies [11,16,26]:

1. Visual analog scale (VAS)—a questionnaire-based scoring system for severity of drooling [11].
2. Drooling quotient—a direct semiquantitative observation [26].
3. Drooling severity and drooling frequency—a questionnaire-based scoring system [16].

Visual Analog Scale (Scale of 0-10). VAS is a semiquantitative scale marked by the chief caretaker who was not told the exact decimal point of the ruler. It was measured before treatment and then daily during TAC treatment course until the end of the course. The chief caretaker put a mark on the scale from 0-10 (Fig 1), with the severity of drooling scaled

Table 1. Drooling score

Drooling Severity		
Score	**Grade**	**Description**
1	Dry	Never drools
2	Mild	Only lips wet
3	Moderate	Wet on lips and chin
4	Severe	Drools to extent that clothing becomes damp
5	Profuse	Clothing, hands, tray, and objects become wet

Drooling Frequency	
Score	**Description**
1	Never drools
2	Occasionally drools
3	Frequently drools
4	Constantly drools

Drooling score (range = 2-9) = drooling severity (range = 1-5) + drooling frequency (range = 1-4).

as 10 meaning very severe and 0 meaning no drooling. The scale of 10 cm was then assessed and scored by a research assistant blinded to the time course of treatment.

Drooling Quotient. We applied the direct observation of drooling episodes using the drooling quotient on a daily basis to avoid the daily fluctuation. We use the same time slot every day to improve the standardization. The actual episode of drooling was observed by a research assistant and scored during two periods of 15 minutes each, separated by a 60-minute interval. The children were evaluated at least 1 hour after lunch, while awake and sitting erect. For those patients who could not sit erect, they were allowed to sit on their mothers' laps. The presence or absence of drooling was assessed every 15 seconds over a 15-minute period (total = 4 × 15 = 60 observations). An episode of drooling was defined as any new saliva leaving the chin.

Drooling quotient was expressed as the ratio of the observed drooling episodes divided by the total number of observations, which is a percentage (drooling quotient = number of drooling episodes/60 × 100%). A research assistant, blinded to the course of the treatment, assessed drooling quotient. The mean of two drooling quotients on the same day before TAC and the mean of two drooling quotients on the last day of TAC treatment was scored to assess the efficacy of TAC.

Drooling Severity, Drooling Frequency, and Drooling Score. A questionnaire-based semiquantitative assessment was given to the chief caretaker, who recorded the severity and frequency of drooling based on a validated scale before and after TAC. Drooling score for both the drooling severity and drooling frequency were added together to make a combined drooling score from 2-9 (Table 1). Thus, a child with drooling severity rated mild (rank = 2) and drooling frequency rated frequent (rank = 3) would have a combined drooling score of 5, or 2 + 3.

All three outcome measures were recorded before and after a course of TAC. Successful treatment with TAC was defined by any positive change (i.e., decrease) in VAS, drooling quotient, drooling severity/drooling frequency (i.e., drooling score).

Statistical Analysis

The VAS, mean of two drooling quotients, and drooling score (drooling severity + drooling frequency) before TAC were obtained as pretreatment data. The VAS and mean of two drooling quotients and drooling score after TAC were obtained as the post-treatment data. Pre- and post-treatment data were analyzed using Wilcoxon matched-pairs signed-rank analysis.

Ethical Consideration

This study was approved by the Ethics Committee of the Faculty of Medicine of the University of Hong Kong. The parents were informed about the methodology and written consent was obtained.

Results

The drooling database consisted of 10 children (five males and five females). The mean age of the cohort is 7.3 years old (median = 5.5; S.D. = 4.762; range = 2-18 years of age).

Drooling Database

The general factors contributing to drooling difficulties were collected as a drooling database for the cohort (Table 2). Patient 1 had meningitis at 5 months. Patients 4, 5, and 8 suffered from Angelmann Syndrome. Patient 2 had Trisomy 9p. Patients 3, 6, 7, 9, and 10 had neurologic sequelae because of neonatal hypoxic-ischemic encephalopathy. Patients 1, 3, 9, and 10 suffered from cerebral palsy; Patient 1 was a mild cerebral palsy patient, meaning an independent walker (spastic diplegia), and Patients 3, 9, and 10 were severe cerebral palsy patients (no independent mobility and all had spastic tetraplegia). Eight children had epilepsy and were on antiepileptic drugs. None of these children had been receiving clonazepam, which is well known in creating drooling problems as one of its side effects.

Statistically significant improvement in all three outcome measures was seen (Table 3). The mean VAS decrease from 6.6 (pre-TAC) to 4.67 (post-TAC) (P = 0.002) (Fig 2). The mean drooling quotient decreased from 14.016% (pre-TAC) to 8.335% (post-TAC) (P = 0.0078) (Fig 3). The mean drooling score also decreased form 7.4 (pre-TAC) to 4.4 (post-TAC) (P = 0.002) (Fig 4).

After reviewing the daily diary, the onset of effect of TAC in decreasing drooling frequency or severity was observed within three to five sessions. The effect was sustained over the course of 30 sessions and the efficacy was noted with increase in the number of sessions. The mean follow-up of these patients was 6 months. According to the mothers, improvement of drooling persisted in most children, except during episodes of febrile illnesses.

Side Effects

None of the children developed any side effects. Initial crying from fear and possible minor pain occurred in the first few sessions. However, most children adapted easily and tolerated the technique well. There was no default rate.

Table 2. General factors of ten children with intractable drooling

Code	Sex	Age	Primary Diagnosis	Phase of Illness	Intelligence Level	Parental Occupation	Oral-motor Dysfunction Measure of Severity	Eating Method/ Ability	Positioning Information	Severity of Drooling*	Nutritional Status	Dental Status; Medications	Other Associated Medical Problems
1	M	18y	CP, MR	Chronic	MMR	U	HPT	Self fed	W	Moderate	N	N; AED	Epilepsy
2	M	3y	MR	Chronic	SMR	MA	LFT	Fed by caretakers	B	Severe	N	N; AED	Epilepsy
3	M	2y	CP, MR	Chronic	SMR	MA	HPT	Fed by caretakers	S	Moderate	N	N	—
4	M	12y	MR	Chronic	SMR	MN	LFT	Self-fed	W	Moderate	N	N; AED	Epilepsy
5	M	9y	MR	Chronic	SMR	MN	LFT	Self-fed	W	Moderate	N	N; AED	Epilepsy
6	F	5y	MR	Chronic	SMR	MN	HPT	Fed by caretakers	S	Moderate	N	N	—
7	F	5y	MR	Chronic	SMR	MN	HPT	Fed by caretakers	B	Severe	N	N; AED	Epilepsy
8	F	6y	MR	Chronic	SMR	MN	LFT	Fed by caretakers	W	Moderate	N	N; AED	Epilepsy
9	F	5y	CP, MR	Chronic	SMR	MN	HPT	Fed by caretakers	B	Severe	N	N; AED	Epilepsy
10	F	8y	CP, MR	Chronic	SMR	MN	HPT	Fed by caretakers	B	Severe	N	N; AED	Epilepsy

* Severity of drooling = quantitative measure (objective) and perceived social and functional impairment (subjective).

Abbreviations:
AED = Antiepileptic drugs
B = Bedridden
CP = Cerebral palsy
HPT = Hypertonic; inability to close lip at rest, poor tongue movement
LFT = Low facial tone, inability to close lip at rest, poor tongue movement
MA = Managerial
MMR = Moderate mental retardation
MN = Manual
MR = Mental retardation
N = Normal
S = Sitter
SMR = Severe mental retardation
U = Unemployed
W = Walker

Discussion

We have demonstrated the efficacy and safety of tongue acupuncture as an adjunctive treatment in children with severe drooling. Our cohort was unable to comply with oral-motor stimulation or behavior modification program. Surgical treatment for drooling problem was not easily accepted in this Chinese community. Thus TAC may be an alternative option for children with intractable drooling problem.

The tongue is a very important organ within the oral cavity. It facilitates biting, swallowing, taste, speech, and emotion through local contact by kisses. In over two millennia of traditional Chines medicine, clinical diagnosis relies on the following four essential criteria: observation, "tongue diagnostics," history taking, and pulse diagnostics. Thus the tongue is the window for diagnosing diseases in traditional Chinese medicine. The tongue is rich in neural (glossopharyngeal and vagus nerves), vascular, and lymphatic supply. Thus by single and repeated local deep stimulation of the tongue using acupuncture to acupoints, the key and core center for linking the vascular-lymphatic-neural networking might be ignited. The physiologic basis of efficacy of TAC in terms of immediate, intermediate, and long-term effects might be explained by resignalling and potentiation of neural receptors through repeated direct stimulation [27].

In a study of cerebral palsy patients with oral involvement using orofacial electromyography and swallowing frequencies, it was reported that drooling was caused by an inefficient and inadequate swallowing mechanism [2]. The pathophysiologic basis of TAC in improving drooling is unknown. With our clinical experience in stroke patients

Table 3. Outcome measures for efficacy of tongue acupuncture

	Pre-TAC			Post-TAC			Wilcoxon Match-pairs Signed-rank Test P Value
	Mean	Median	SD	Mean	Median	SD	
Visual analog scale	6.6	6.9	2.274	4.67	4.5	2.111	0.002
Drooling quotient	14.016	12.080	5.839	8.335	6.252	6.311	0.0078
Drooling score	7.4	7	0.9661	4.4	4	0.5164	0.002
Drooling severity	4.3	4	0.483	2.4	2	0.516	0.002
Drooling frequency	3.1	4	0.5675	2	2	—	0.0001

Abbreviations:
SD = Standard deviation
TAC = Tongue acupuncture

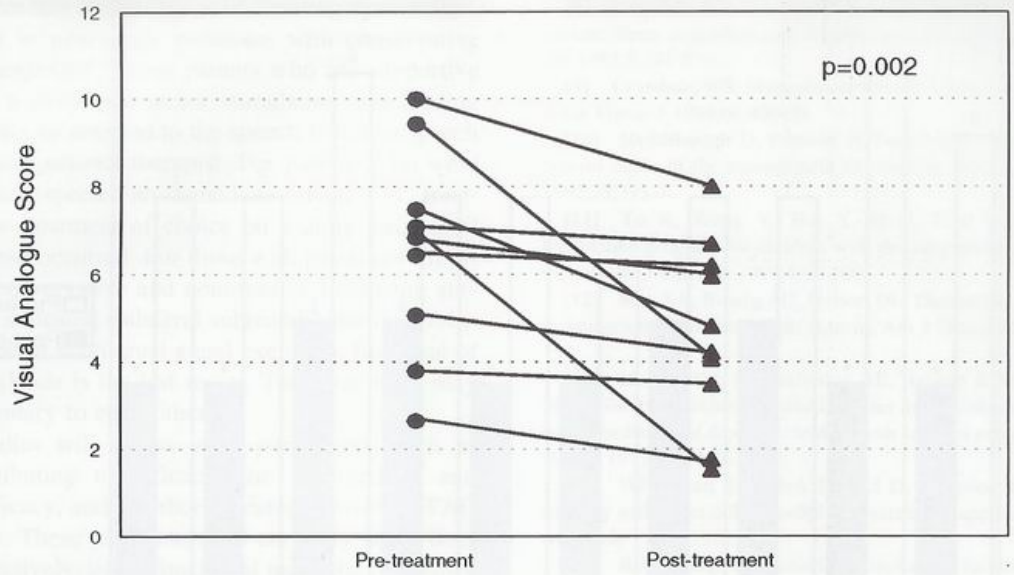

Figure 2. Visual analog scale.

with pseudobulbar palsy, we discovered that there were specific acupoints on the tongue that affect the swallowing mechanism [31,33]. A hypothesis that TAC stimulated the rich neural network in the tongue, which are connected to salivary glands and tongue muscles via cranial nuclei in the brainstem, and improves salivary secretion and swallowing mechanism, respectively, was postulated.

Anecdotal clinical experience for patients with learning disability and ataxia also revealed that there were specific acupoints on the tongue to improve cognitive function and ataxia, respectively. Improvement in alertness, attention, and emotional state of a child through "starting-up" with TAC to acupoints rich in neural networking to the frontal cortex through the brainstem and cerebellum may explain the improvement in the child's awareness of drooling. Thus by a mechanism of re-networking the "brain systems," plasticity of the brain is evident in various functional disabilities [28-34].

With the availability of techniques to address the issue of the mechanics of normal swallowing, future TAC

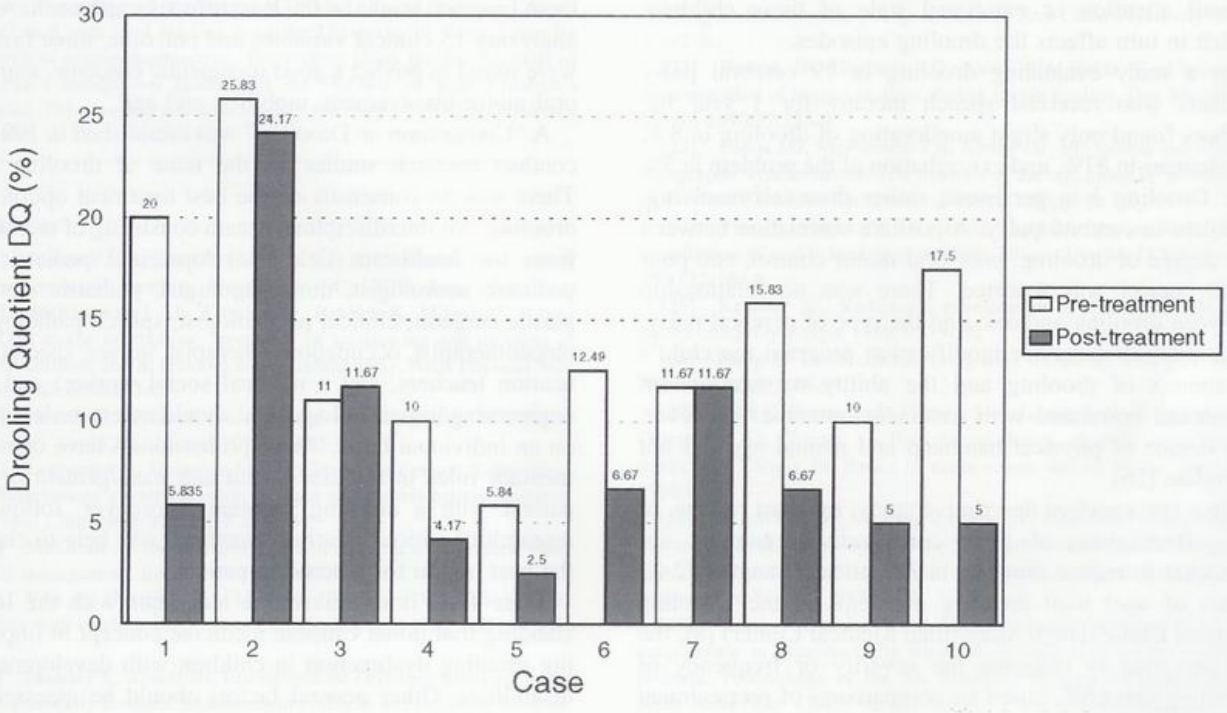

Figure 3. Drooling quotient.

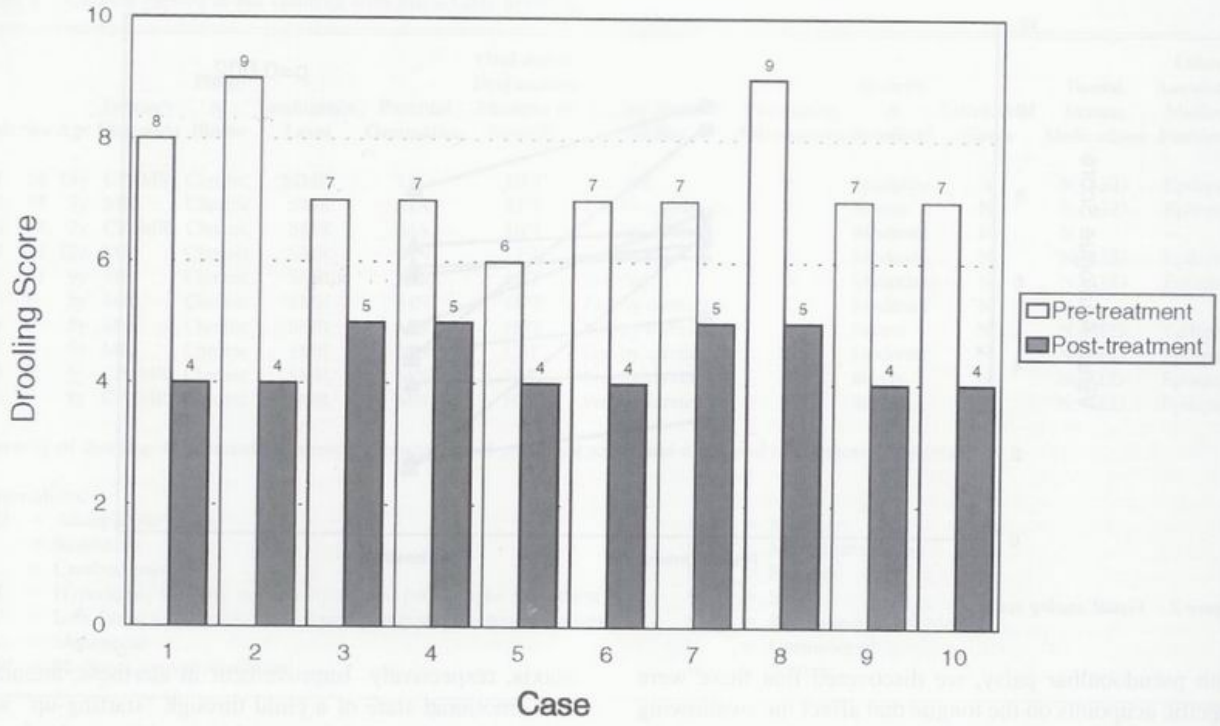

Figure 4. Drooling scores.

research should be aligned with these noninvasive investigations to document the reason for improvement, for example, video fluoroscopy swallowing study (VFSS) to assess swallowing mechanism in cooperative children. Assessing the attention and cognitive ability of these children is underway to see whether TAC improves the overall attention or emotional state of these children, which in turn affects the drooling episodes.

In a study evaluating drooling in 79 cerebral palsy patients who received speech therapy for 1 year the authors found only slight amelioration of drooling in 8%, no changes in 87%, and exacerbation of the problem in 5% [6]. Drooling is a persistent, rather than self-resolving, problem in cerebral palsy. A positive correlation between the degree of drooling, poor oral-motor control, and poor head control was reported. There was no relationship between drooling and sex, and the type of cerebral palsy. In a study of behavior modification program the child's awareness of drooling and the ability to swallow on command correlated with treatment outcome. However, the degree of physical handicap and mental age did not correlate [26].

In a retrospective descriptive study, by chart review, of the effectiveness of three conservative treatment approaches to reduce drooling in 36 patients (range = 2-23 years of age) with drooling problems at the Drooling Control Clinic (Hugh MacMillan Medical Center) [8], the success rate in reducing the severity or frequency of drooling was 66%, based on comparisons of pretreatment and posttreatment drooling rank (drooling severity + drooling frequency) ($P < 0.001$) The mean change in drooling rank was 1.8 points (S.D. = 1.9) [35]. The three conservative approaches consisted of the following treatments: (1) no direct intervention; (2) feeding/oral stimulation program; and (3) behavior modification program. The study suggested that feeding/oral stimulation treatment program might be the least effective approach. After analyzing 15 clinical variables and outcome, three factors were found to predict a good therapeutic outcome, namely oral motor involvement, mobility, and age.

A "Consortium on Drooling" was established in 1991 to conduct research studies on the issue of drooling [1]. There was no consensus on the best treatment option for drooling. An interdisciplinary team consisting of members from the healthcare field (developmental pediatrician, pediatric neurologist, otolaryngologist, pediatric dentist, plastic surgeon, clinical psychologist, speech pathologist, physiotherapist, occupational therapist, nurses, special education teachers, and a medical social worker) and the engineering/biotechnology field should assess each patient on an individual basis. These professionals have complementary roles in the assessment and management of the patient with a drooling problem. Moreover, follow-up research of various treatment methods will help to clarify the best option for a drooling patient.

Thus TAC is an innovative technique with the long-standing traditional Chinese medicine concept in improving drooling dysfunction in children with developmental disabilities. Other general factors should be assessed to streamline the drooling patients to various groups of

treatment according to the degree of drooling dysfunction. Improvement in neurologic problems with conservative treatment is expected. Those parents who are supportive and perform a daily oral motor stimulation and feeding program should be referred to the speech therapist/speech pathologist/occupational therapist. For parents who were supportive and expected an immediate effect, TAC treatment was the treatment of choice on a daily basis until possible relapse occurred. For those with persistent drooling, despite conservative and noninvasive treatment, surgery may be indicated (bilateral submandibular duct relocation/excision or sublingual gland excision). Removal of the salivary glands is the last resort. These methods may be complementary to each other.

Future studies will assess other parameters, such as factors contributing to efficacy, the intermediate and long-term efficacy, and whether repeated courses of TAC are necessary. These parameters are currently being documented objectively, using functional magnetic resonance imaging and positron emission tomography, in which the function of specific acupoints in normal subjects proves the philosophy of traditional Chinese medicine.

TAC is an innovative technique invented after more than 20 years of clinical experience in humans, and this technique will be disseminated to medical professionals after mapping out the acupoints on the tongue using objective criteria. TAC research is currently being conducted with standardized methodology after objective documentation and pilot studies, such as a randomized placebo-controlled trial using sham acupuncture.

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