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Oral placement therapy: A novel approach for addressing lead metal toxicity in autistic children

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***Abstract*---**Objective: To investigate the effect of OPT (Oral Placement Therapy) on Blood lead levels (BLL) measured in microgram/dl and CARS 2 ST (Childhood Autistic Rating Scale 2nd edition Standard version) scores of autistic children both in the short and long term. It also studied the clinical views in speech, feeding and sensory needs by using OPT to bring down the BLL along with levels of CARS2-ST scores in children under autism spectrum disorder. Methods: This comparative observational study was conducted from April 2019 to January 2020, used repeated measures design involving 40 children under autism spectrum aged between 3 to 8 years with concerns of mouthing and smelling of inedible things. 40 participants were recruited to understand the performance benefits of a 5 months OPT versus 10 months OPT on severity of autism as measured by the CARS2-ST from 5 months to 10 months. An asymp sign test was used to compare the differences in CARS2-ST scores in the two repeat measures.Results: Wilcoxon signed-rank test showed statistically significant median decrease in BLL (7.3) and (6.8) at 5 and 10 months respectively when compared before therapy (7.75). Similar results were seen for CARS2ST. Median values for CARS2ST at 0, 5 and 10

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months were (33.5), (33) and (31.25) respectively. Conclusion: It provides evidence that OPT can be used objectively to improve the BLL and CARS2-ST scores in autistic children. This study also suggests that OPT should be continued longer despite the initial positive effect so that CARS2-ST and BLL keeps improving. However, before generalizing the findings of the study, more research is needed on OPT and its impact on CARS2-ST and BLL.

***Keywords*---**oral placement therapy, childhood autistic rating scale, blood lead levels, autism spectrum disorder.

Introduction

Sensory processing disorder also known as Sensory Integration Dysfunction, first identified in 1960's, is a neurological disorder causing difficulties with processing and responding to sensory stimuli from environment and from within an individual's own body[1]. Children with autism may have atypical responsivity to sensory stimuli as high as 95%[2]. Individuals with autism spectrum disorders monitor impaired sensory modulation (sensory seeking, sensory avoidance), aversion to oral care and absence of oral exploratory phase, sensitivity to taste, texture, smell, sight of food. They always seem to have something in their mouths like toys, pens, pencil tips, gum, candy, or inedible objects (i.e., paper clips, rubber bands, shirt sleeves and collars, cosmetics, leather, chalk pieces thereby increasing the risk of exposure to lead compared to neurotypical children. They may also stuff food, instead of chewing, throw food, spit or vomit, avoid utensils or food to lips. Lead poisoning considered to be one of the most common causes of neurodevelopmental impairment in childhood, has deleterious effects on the development of widespread brain areas including those implicated in cognitive, communication, and social functioning[3]. In several cases, a temporal association was noted between elevated blood lead levels and the emergence of autistic symptoms.[4]

The empirical evidence[5-9] suggests that 70% of all children diagnosed with autism have sensory processing disorders (SPD), confirming the existence of sensory and motor difficulties for many children with autism at some point in their early development. The atypical eating behaviours of autistic children, along with habitual mouthing and pica, make it hard to determine whether increased lead levels are a cause or a consequence of autism.[10] Since OPT involves the holistic approach for addressing the oromotor sensory concerns, this study was designed to address the lead metal toxicity in Indian sample of autistic children using Oral placement therapy as a novel approach.

Autism Spectrum Disorder (ASD) is a developmental disorder pertaining to children with characteristic of communication issues, social interactions, restricted, repetitive behavior that impair social, occupational and daily functioning[11] Individual with ASD varies widely in terms of behavior, intelligence and abilities, symptoms often include delay in speech, understanding and use of language, relating difficulties with objects, events and people, unusual play with objects and toys, difficult coping with routine change, repetitive movements and

behavior.[12]

Studies on oral health and ASD children are limited with few evidence-based research with multiple difference of opinion showing oral health of children with and without disabilities. Hypersensitivity in ASD children to sensory input can also interfere with mouthing issues and oral care, such features make it difficult for parents to care for their children's oral hygiene. Furthermore, children with ASD are often impaired in responding to social cues and communication, as high as 40 percent of children with ASD do not talk at all.[13,14]

Sara Rosenfeld-Johnson a Speech Language therapist in America found that speech and feeding are interrelated and difficult to separate. In 1973 she developed her own technique in conjunction with OTs (Occupational Therapy) and Physios, which is called the OPT (Oral Placement Therapy). For this study, the Talk Tools OPT is used which is one specific type of Oral Motor Therapy. This OPT is not meant to replace other therapy approaches and is often used for motor and sensory difficulties before any other approaches on speech and feeding is introduced.[15-17]

Oral motor therapy is not new and has been documented with many contributions over years. According to Van Riper, the OMT techniques are helpful to facilitate lips, jaw, tongue movement, phoneme sensitivity and production and have been present for centuries now.[18,19] Charles Van Riper or also known as the 'father of traditional articulation therapy' published some of his works on speech correction and describes many lip, jaw and tongue techniques for facilitation and importance of tongue movement for speech skills that aid in retracting, protruding, grooving and curling up of tongue which is important step towards speech facilitation.

He wrote "When the stimulation method fails , they (phonetic placement methods/objects) must be used'.[19-21] Leading specialist such as Van Riper, (1954), The Feedback Model (Mysak, 1971), Young and Hawk, (1955), Green, Moore & Reilly, 2000 and Marshalla (2007), all mentioned the use of objects and tools for articulation therapy and it is grouped by studies as nonspeech oral motor exercises (NSOME) and nonspeech oral motor treatments (NSOMT). Tons of debates on use of tools from practitioner, clinicians and retailers suggest of no vital proof that document any objects or tools to help in learning of speech and Language. The OMT (Oral Motor Therapy) and OPT, a section of OMT is all based on a common agenda to facilitate speech movement with therapy tool assistance, tactile-kinesthetic facilitation technique, and use available oral speech device.

Term of "Oral Placement Disorder" (OPD) was first conceived by Diane Bahr and Sara Rosenfeld-Johnson in 2010. Then OPT was formulated by Talk Tools which represents facilitating techniques by using a needed tactile and proprioceptive treatment after taking a detail understanding and view of extra oral and intraoral structures movement of mouth and thereby get phonetic production for speech and language development. OPT approach are multifold as it uses not just OPT methods alone but may combine with other traditional articulation techniques to bring out sound with speech, often resonated and produced better with repetition and practice. Whichever approach of OPT is facilitated it is important to first

evaluate and assess if the child can make sounds under instruction, if child can produce targeted sounds, then a tactile – proprioceptive placement is not required, and typical style of speech production can begin. If the child cannot attain speech sounds with help of audio- visual input then a thorough assessment of motor functions and oral sensory is required, once abnormal oral placements are identified a targeted movement of hierarchy is then used. OPT is practiced until the child can perform sound and speech without tools and facilitation of any kind, and once the placement is attained, it is transitioned into speech. Gregory Lofs and Marshalla have both mentioned that Von Riper placement method is not a NSOME and is an extension and may be just a placement therapy used for phonation.[21-26]

There always remains a debate if heavy metal toxicity is the cause or consequence of Autism. Exposure to lead is a major Public Health concern globally, and studies have reported an alarming rise of ASD children over the years with possible exposure with lead and having increase Blood Lead Levels (BLL). Cadmium, arsenic, lead, and mercury have been linked to autism, attention deficit disorder, mental retardation, and death of children.[27] Lead exposure is an insidious problem, causing subtle effects in children at low exposure levels where clinical signs are not apparent.[28] Exposure to environmental levels of lead (Pb) and manganese (Mn) has been associated with detrimental effects to neurodevelopment.[29]

Studies suggest of detoxifying and stop all possible risks that make these children more susceptible to contamination to lead. Some of the exercises such as tactile stimulation and tongue exercises help in possible feeding, mouthing and oromotor issues by increasing oral and tactile awareness. Our study suggests a need for blood lead levels check for every ASD diagnosed child with oromotor sensory issues, as it can help the professionals dealing with autistic children to decide on OPT for the child at an early age and help in reducing BLL and simultaneously even the CARS2-ST Scores.

Objectives

a. To apply Oral Placement therapy to help in speech, feeding and sensory issues in children under the Autism Spectrum.
b. To see the effect after 5 months on BLL and CARS2-ST scores.
c. To see the effect after 10 months on BLL and CARS2-ST scores

The aim of this observational study is to see the effect of 10 months Oral-placement therapy on motor and sensory issues and describe its follow-on effect on blood lead levels and CARS2-ST score of children age 3 to 8 years under Autism Spectrum.

Study definitions

OPT: Oral placement therapy
CARS2-ST: Childhood Autistic Rating Scale-2nd edition –Standard Version
ASD: Autism spectrum disorder
BLL: Blood lead levels measured in microgram per decilitre(ug/dl)

Methodology

Study Design and Participants

This study conducted from April 5, 2019 to January 10, 2020 is based on the observation of children who were residents of Delhi and matched the eligibility criteria. (ethical clearance no: IEC/IDS/38/2019)

Inclusion Criteria in the Study

1. Age group: 3 to 8 years
2. Children under autism spectrum as diagnosed by clinical psychologist/Paediatrician/ developmental paediatrician
3. Children having oromotor concerns with mouthing and smelling issues

Exclusion Criteria

1. Children having any major illness or Systemic diseases.
2. Mouthing issues due to iron deficiency anaemia

The Wilcoxon signed rank non-parametric test was conducted to find out non-normality in the data. It was used to compare two sets of CARS2-ST Scores and BLL that come from the same participants over repeated measures in 5 months and 10 months post OPT.

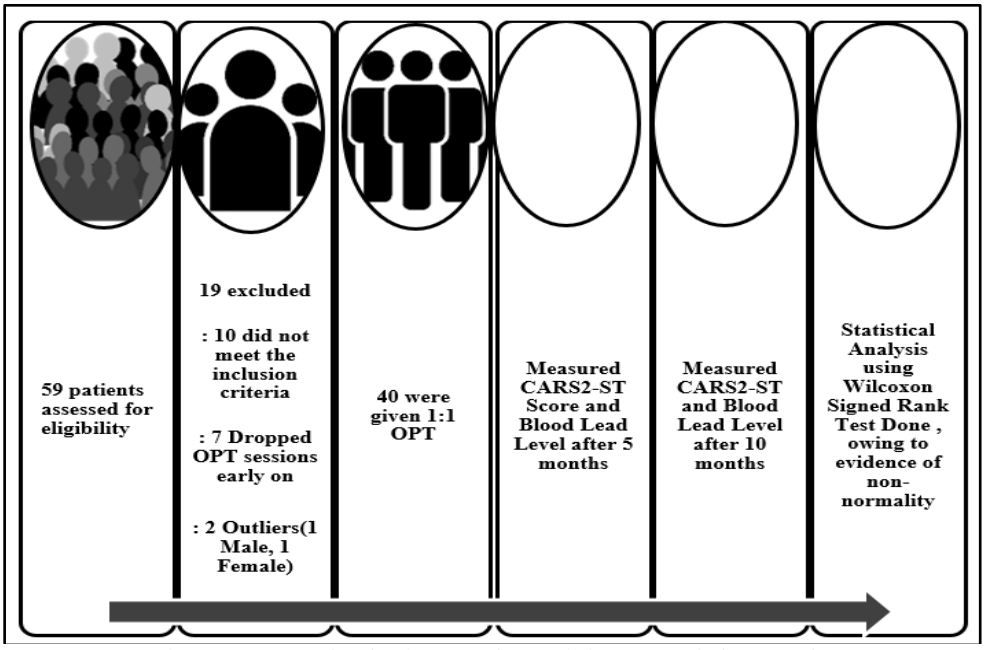


Figure 1: Study design and participants of the study

Study Procedure

Parents coming to the clinic were first asked to fill an enquiry form collecting the background information of the child including diagnosis if made previously, present concerns, ongoing therapies, medications, antenatal, birth history and diet history. If no confirmatory diagnosis of the child was present, a fresh assessment with confirmatory diagnosis was made by using CARS2-ST scores by developmental paediatrician). Following that, parents were interviewed and physical assessment of the child using informal oromotor assessment was done by an Autism Specialist certified in OPT. Study was introduced, after attaining informed consent from parents/caregiver, whose children had oromotor issues with concerns of regular mouthing and smelling of non-edible objects.

At the baseline of study along with the CARS2-ST scores, blood lead levels were also determined to check the lead levels (Blood Lead levels-BLL) before OPT inclusion. Depending upon the severity of oromotor issues including mouthing and smelling they were called for therapies maximum of thrice a week on all 6 working days of the week (Tuesday to Sunday). OPT (Oral placement therapy) was done using Talk-Tools Kit and other tools targeting sensory, feeding and speech issues which was targeted at 1:1 session of 30 minutes.

Parents were given a hands-on home-based oromotor exercise which were to be done at home. Looking at financial constraints, option of buying essential tools were suggested and option of using clinical tools were given, taking utmost care for sanitization and sterilization for each participant. At the end of the 5 months and 10 months of OPT, oromotor assessment was done and BLL taken to see the improvement in oromotor sensory issues and CARS2-ST scores. OPT intervention strategies used in the study have been shown in Table1

Table 1: OPT Intervention Strategies

Oromotor exercises	Oral sensory exercises	Systematic Desensitization through play
Usage of chewy tubes to improve the chewing skills and Bite Blocks for jaw strength	Tactile stimulation on and around lips to improve the awareness/ desensitization	Play face touch game with rubber dolls, Thera putty, playdough, soft toys. Kiss child's face and/or let him kiss parents face. (Only with parents)
Tongue Lateralizers and Elevator tools, Resistance exercises, tongue movement exercises were used for tongue movements and coordination	Icing to improve the oral stimulation and awareness. Brushing of lips and gums to improve the tactile awareness.	Playful taking turns through touching. Wipe the face with warm clothes apply.
Blowing bubbles and horn Kit, straw sucking, Button Pull and lip exercises for lip	Vibratory stimulation to tongue, lips, cheeks.	Food Play to increase new food acceptance.

strengthening.		

Statistical Analysis

A prespecified analysis plan governed all analyses, unless identified. Wilcoxon Signed Rank Test was chosen, owing to evidence of non-normality. Following steps on SPSS 23.0 was used
Analyze > Nonparametric Tests > Legacy Dialogs > 2 Related Samples for all the six analyses. Descriptive statistics summarized participants demographics, pre and post intervention OPT CARS2-ST scores.

Results

In Table 2 the socio demographic characteristics of study participants as shows that out of 40 children participating in the study, the 67.7% were males and 32.5 % were females. Mean age of the children is recorded as mean (5.288) years, SD (1.497) and median age (5.050). Graph 1 shows the age range of the total 40 participants taken for study. The baseline CARST-S2 Scores reads as mean (5.288), median (33.500) and SD (1.497). Graph 2 shows the CARST-S2 Scores before 5 months OPT for total 40 participants in the study. The CARST-S2 Scores before 5 months OPT read as mean (33.773), median (33.500) and SD (2.359). Graph 3 shows the CARST-S2 Scores after 5 months OPT of total 40 participants in the study. The CARST-S2 Scores after 5 months OPT reads as mean (33.170), median (33.000) and SD (2.279). Graph 4 shows the CARST-S2 Scores after 10 months OPT of total 40 participants in the study. The CARST-S2 Scores after 10 months OPT reads as mean (31.193), median (31.250) and SD (2.772). Graph 5 shows the Baseline Blood Lead Level values before OPT of total 40 participants in the study. The Blood Lead Level values before OPT reads as mean (7.743), median (7.75) and SD (0.885).

Graph 6 shows the Blood Lead Level values after 5 months of OPT for total 40 participants in the study. The Blood Lead Level values after 5 months of OPT reads as mean (7.340), median (7.300) and SD (0.861). Graph 7 shows the Blood Lead Level values after 10 months of OPT for total 40 participants in the study. The Blood Lead Levels values after 10 months of OPT as mean (6.898), median (6.800) and SD (0.856). Outcomes were the CARS2-ST Scores for ASD and Blood Lead Levels in response to OPT treatment which were measured before the OPT and post 5 months and 10 months OPT. 40 participants were recruited to understand the performance benefits of a 5 months OPT versus 10 months OPT on severity of autism as measured by the CARS2-ST from 5 months to 10 months. An asymp sign test was used to compare the differences in CARS2-ST scores in the two repeat measures.

a. Wilcoxon signed-rank test determined that there was a statistically significant median decrease in CARS2-ST Score (33) when participants underwent OPT for 5 months compared to before therapy (33.5), $z = - 5.046$, $p = 0.000$.
b. Wilcoxon signed-rank test determined that there was a statistically significant median decrease in CARS2-ST Score (31.25) when participants underwent OPT for 10 months compared to before therapy (33.5), $z = -$

5.536, p = 0.000.

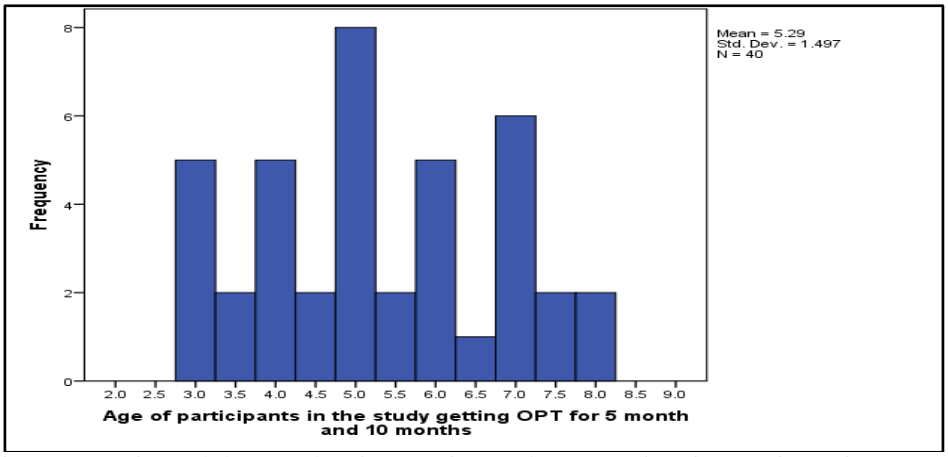
c. Wilcoxon signed-rank test determined that there was a statistically significant median decrease in CARS2-ST Score (31.25) when participants underwent OPT for 10 months compared to 5 months therapy (33), z = - 5.529, p = 0.000.

40 participants were recruited to understand the performance benefits of a 5 months OPT versus 10 months OPT on Blood Lead Levels as measured from 5 months to 10 months. An asymp sign test was used to compare the differences in Blood Lead Levels in the two repeat measures.

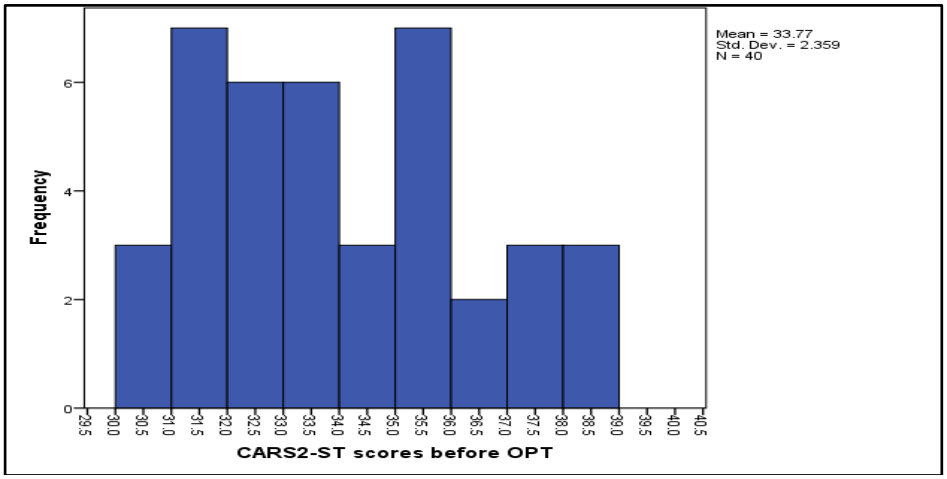
a. Wilcoxon signed-rank test determined that there was a statistically significant median decrease in Blood Lead Levels (7.3) when participants underwent OPT for 5 months compared to before therapy (7.75), z = - 5.320, p = 0.000.
b. Wilcoxon signed-rank test determined that there was a statistically significant median decrease in Blood Lead Levels (6.8) when participants underwent OPT for 10 months compared to before therapy (7.75), z = - 3.672, p = 0.000.
c. Wilcoxon signed-rank test determined that there was a statistically significant median decrease in Blood Lead Levels (6.8) when participants underwent OPT for 10 months compared to for 5 months therapy (7.3), z = - 2.044, p = 0.041.

Table 2: Socio demographic Characteristics with baseline and following OPT scores of CARS2-ST and BLL

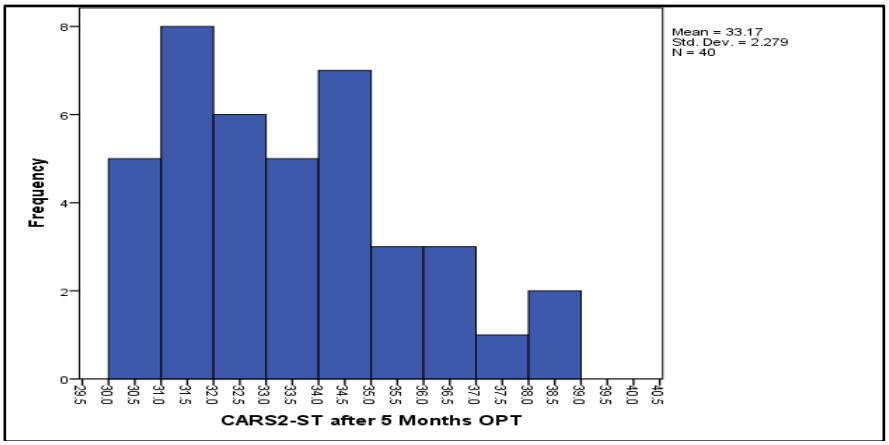
(n= 40)	Mean	Median
Age	5.288	5.050
CARS2-ST before OPT	33.773	33.500
CARS2-ST after 5 months OPT	33.170	33.000
CARS2-ST after 10 months OPT	31.193	31.250
BLL before OPT	7.743	7.750
BLL after 5 months OPT	7.340	7.300
BLL after 10 months OPT	6.898	6.800



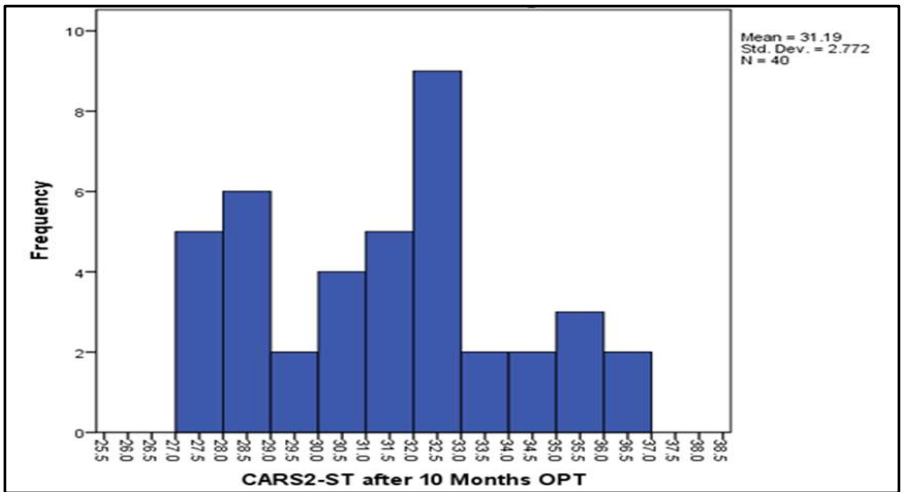
Graph 1. Age of participants in the study getting OPT for 5 month and 10 months.



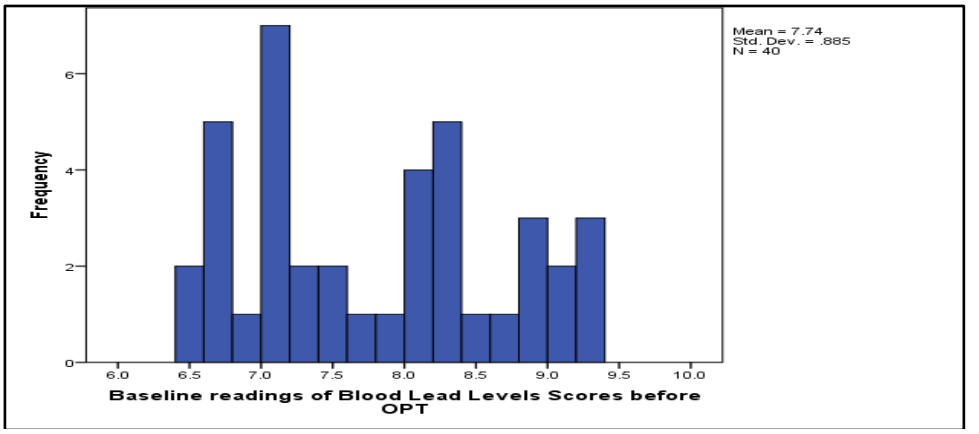
Graph 2: CARST-S2 Scores before OPT



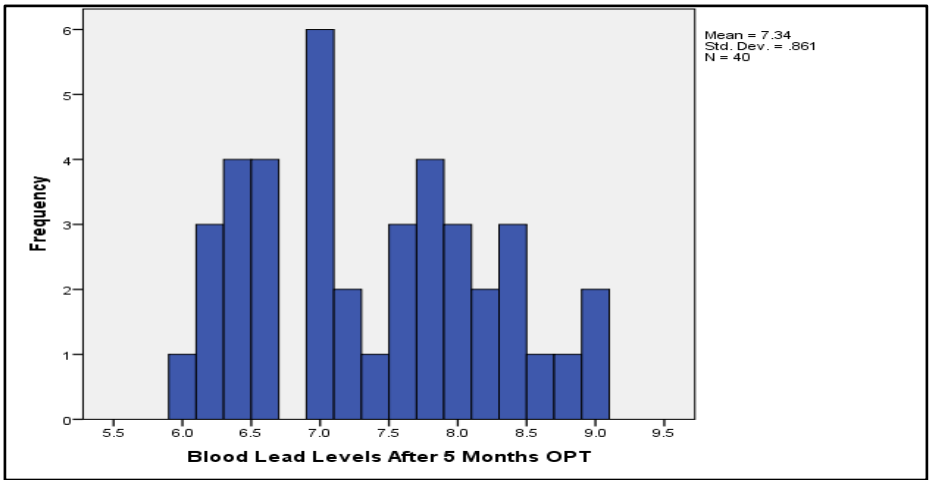
Graph 3: CARST-S2 Scores After 5 months OPT



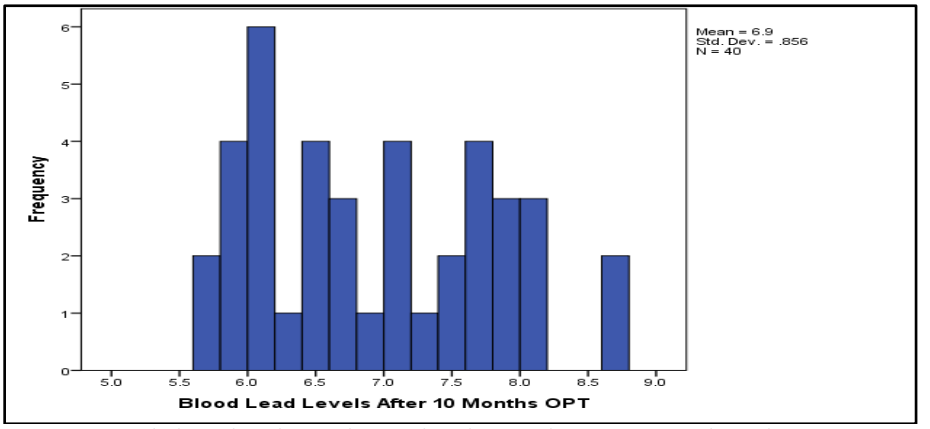
Graph 4: CARS2-ST Scores After 10 months OPT



Graph 5: Baseline readings of Blood Lead Level (BLL)values before OPT



Graph 6: Blood Lead Level values after 5 months of OPT



Graph 7: Blood Lead Level values after 10 months of OPT

Table 3: Wilcoxon Signed Ranks Test: Impact of OPT on CARS and BLL

		N	Mean Rank	Sum of Ranks
CARS2-ST after 5 months OPT -CARS2-ST before OPT	Negative Ranks	32	16.50	528.00
	Positive Ranks	0	.00	.00
	Ties	8		
	Total	40		
CARS2-ST after 10 months OPT - CARS2-ST before OPT	Negative Ranks	40	20.50	820.00
	Positive Ranks	0	.00	.00
	Ties	0		
	Total	40		
CARS2-ST after 10 months OPT - CARS2-ST after 5 months OPT	Negative Ranks	40	20.50	820.00
	Positive Ranks	0	.00	.00
	Ties	0		
	Total	40		
BLL after 5 months OPT - BLL before OPT	Negative Ranks	37	19.00	703.00
	Positive Ranks	0	.00	.00
	Ties	3		
	Total	40		
BLL after 10 months OPT - BLL before OPT	Negative Ranks	30	22.77	683.00
	Positive Ranks	10	13.70	137.00
	Ties	0		
	Total	40		
BLL after 10 months OPT - BLL after 5 months OPT	Negative Ranks	25	18.52	463.00
	Positive Ranks	11	18.45	203.00
	Ties	4		
	Total	40		

Table 4: Wilcoxon Signed Ranks Test based on Positive Ranks

	CARS2-ST after 5 months OPT - CARS2-ST before OPT	CARS2-ST after 10 months OPT - CARS2-ST before OPT	CARS2-ST after 10 months OPT - CARS2-ST after 5 months OPT	BLL after 5 months OPT - BLL before OPT	BLL after 10 months OPT - BLL before OPT	BLL after 10 months OPT - BLL after 5 months OPT
Z	-5.046	-5.536	-5.529	-5.320	-3.672	-2.044
Asymp. Sig. (2-tailed)	.000	.000	.000	.000	.000	.041

Discussion

This study aimed to see the effect of 10 months of Oral Placement Therapy and describe its follow-on effect on blood lead levels and childhood autistic rating scale scores of children age 3 to 8 years under Autism Spectrum. In our study, all the statistical findings had a common outcome which shows that OPT helps in bringing the CARS2-ST scores and BLL down significantly. Wilcoxon signed rank test was used in the study population. In the beginning of the study, CARS2 ST median score was 33.5, which was reduced to 33 after 5 months of OPT, which further reduced to 31.25 after another five months of OPT. This decrease in CARS2ST score from 5 months to 10 months was due to the significant decline in blood lead levels in the study population from 0 to 10 months.

In the first five months in the study population, OPT had mild to moderate effect on oral sensory issues like smelling and mouthing of non-edible things, but due to continuous and vigorous OPT sessions, motivation and compliance by the parents by seeing the initial positive effect of OPT at 5 months, there was a constant and significant decrease of BLL at 10 months of therapy. As a result of improvement in oral sensory system, exposure to lead due to toys, creams, paints, utensils, etc. was reduced in study population and hence there was a decline in blood lead levels from 0 to 10 months. Initial median BLL were 7.75 which were reduced to 7.73 at 5 months which further showed significant decrease in median values of BLL 6.8 at end of 10 months of therapy. Very a few studies have been conducted to assess the effect of oromotor therapy on different variables like behaviors, oral motor and oral sensory issues. etc. and ours is the first study of its kind to assess the effect of oral placement therapy on blood lead levels and childhood autistic rating scale scores in autistic children.

A case study was done by A. K. Aswathy, et.al[30] “Addressing Oral sensory issues in children with autism spectrum disorder”. In this study the effect of Oral motor intervention strategies was studied on oral sensory, articulation and speech. The study reported better tolerance two food texture, improvement in speech intelligibility and articulation but it did not study the effect of blood lead levels on CARS2ST scores. In another study done by Twachtman-Reilly, J et.al[31], nature of

oromotor difficulties especially feeding problems in Autism spectrum children were studied in relation to behavior in school settings. The study did not measure the blood lead levels in this subset of population, where lead could be a possible causative agent causing behavioral problems due to oromotor and feeding difficulties in the sample population.

In accordance with the above two studies, our study also showed positive effects of oromotor therapy on oral motor and oral sensory issues. In addition, our study also showed the positive effect of oral placement therapy (a type of oromotor therapy) on reducing blood lead levels by improving the oral sensory issues (mouthing, smelling of non-edible things) and thereby improving CARS- 2ST scores in the sample population. Our study could provide a possible alternative to chelation therapy in managing blood lead levels at initial stages in autistic children in future. As there always remains a dilemma, if lead metal toxicity is the cause or consequence of Autism, hence it is noteworthy to assess for oro motor issues in every autistic child with appropriate oral placement therapy strategies to reduce the risk of lead toxicity in autism spectrum disorder.

The limitations faced during the study were the non-random and non-normal sampling design which was subject to potential weakness of bias and strength of test. Moreover, the sample may not be sufficient to interpret the actual degree of the findings in totality; as all subjects were allocated from one state/ Union Territory of India i.e., Delhi. However, the study population was chosen to reflect most ASD children aged 3 to 8 years. Finally, this study was moderately small, a larger study is needed. The levels of other environmental pollutants, for example, mercury, aluminum which may work in synergy to enhance lead toxicity were not assessed. Since this study was carried out on a limited participants from Delhi, India, it is suggested the study should be carried out on larger set of target population which may be more conclusive in future. Further, more robust studies are required to generalize the positive effects of oral placement therapy on blood lead levels in autism and its subsequent effect in improving childhood autistic rating scale scores.

Conclusion

This study involving 40 participants with 27 males and 13 females, concluded that in all the statistical findings a common outcome has been found, which shows that OPT helps in bringing the CARS2-ST scores and BLL down significantly. In the first phase of the study in 5 months, shows significant decreases in CARS2-ST scores and BLL. The statistical findings indicate that continued 10 months of OPT therapy has continued benefits which were also very significant as comparable to the initial 5 months, and it suggests that OPT should be continued despite the initial positive effect so that in ASD child CARS2-ST and BLL keeps improving. However, for better results, a suggestive of more rigorous and extensive therapy is needed for 10 months minimum and above.

Ethical clearance

It was taken from the Institutional Ethics Committee (IEC) of Institute of Dental Sciences, Uttar Pradesh, India.

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