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## Factors associated with cooperative levels of Autism Spectrum Disorder children during dental treatments

### ABSTRACT

**Aims** To evaluate the cooperation rates in Thai ASD patients and analyse factors associated with cooperation levels in ASD patients during dental treatment.

**Materials and methods** A group of 95 subjects aged between 3–18 years diagnosed with ASD and who had been evaluated for sensory processing by an occupational therapist received dental treatment in the dental clinic at the Rajanukul Institute of Bangkok (Thailand), which is the governmental institute providing medical and dental services to the community, including special needs children. Data were collected from medical records and parental questionnaires as well as behaviour rating during dental treatment using the Frankl rating scale. Qualitative data were analysed using descriptive analysis. Chi-square test and multiple logistic regression analysis were used to analyse the factors associated with clinical behaviour during dental

treatment ( $p < 0.05$ ).

**Results** Total subjects were 95 (82 males, 13 females). Mean age was  $8.7 \pm 3.9$  yrs. Age, behaviour before dental treatment and dental procedures ( $p < 0.001$ ), proprioception system ( $p = 0.021$ ), tactile system ( $p = 0.043$ ), education ( $p = 0.004$ ), verbal communication ability ( $p = 0.007$ ), ability to follow the instruction ( $p = 0.014$ ), tooth brushing habit ( $p = 0.014$ ), and toilet training ( $p = 0.001$ ) were significantly associated with a cooperative behaviour during dental treatment. Multiple logistic regression analysis showed that age, education, and behaviour before dental treatment were associated with the cooperative behaviour of ASD patients during dental treatment. The level of cooperation in ASD dental patients aged 11–18 yrs was 11 times higher than for patients aged 3–7 yrs old (adjusted OR=11.51,  $p = 0.004$ ). Subjects who had attended special education system showed a cooperation level almost 13 times greater than subjects who did not (adjusted OR=12.7,  $p = 0.014$ ). Subjects who showed a positive behaviour before the dental procedure started showed higher cooperation during dental treatment, 15 times greater than that of patients with negative behaviour (adjusted OR=15.26,  $p < 0.001$ ).

**Conclusion** School-age ASD children who have been trained in special education programmes and showed positive behaviour before dental treatment show a higher cooperation level during treatment.

**Keywords** Autism Spectrum Disorder; Cooperation; Dental treatment.

### Introduction

Autism spectrum disorder (ASD) is a persistent neurodevelopmental condition with early childhood onset. Latest report from the Centers for Diseases Control and Prevention (CDC) showed that the overall prevalence of ASD continued to rise. In 2010, the estimated prevalence of ASD in the United States was 14.7 per 1,000 [CDC, 2014]. The American Academy of Paediatric Dentistry (AAPD) reported that the prevalence of ASD in children aged 3–17 years was 1 in 91 [Kogan et al., 2009]. A study in the Republic of Korea in 7–12 years old children showed a prevalence of 1 per 38 [Kim et al., 2011]. In Thailand, the prevalence of autistic children was 9.9 per 10,000 [Poolsuppasit et al., 2005].

ASD patients are frequently found with social impairment, language impairment, and repetitive or stereotyped behaviours. Some might be suspected of self-harming behaviours, aggression, temper tantrums as well as mental impairment, convulsions, and

sensory integrating problems [Friedlander et al., 2006; American Psychiatric Association, 2013]. Communication impairment, atypically response to visual, auditory, tactile, olfactory, or gustatory stimuli results in the difficulties when it comes to delivering dental treatment [Watling et al., 2007; Stein et al., 2014]. Some ASD patients have average cognitive function and are able to receive treatment in a regular dental clinic setting with some behavioural management techniques, while others may be difficult to treat without pharmacological management [Klein et al., 1998; Limeres-Posse et al., 2014]. Previous studies showed that a significantly higher percentage of ASD patients were uncooperative during dental treatment compared to the unaffected group [Loo et al., 2008; Loo et al., 2009; Marshall et al., 2007]. Some studies have analysed the factors associated with cooperation levels of ASD patients during dental treatment. They found that age, concurrent diagnosis, behavioural problems, and patient's life skills were associated with a cooperative behaviour while gender, use of psychotropic medication and treatment experience were not associated [Loo et al., 2009; Marshall et al., 2007; Dinnissen et al., 2015; Wink et al., 2015]. Marshall and colleagues [2007] used parent surveys and parent interviews to analyse the association between the factors related to levels of cooperation during dental treatment of ASD patients. They found that multiple factors were associated with levels of cooperation during dental treatment: appointment type, concurrent diagnosis, communication difficulties, the ability to follow instructions, learning ability, tooth brushing, toilet training, and the ability to sit for a haircut. By reviewing dental charts, Loo and colleagues [2009] found three characteristics associated with patient cooperation in dental clinics: age, ASD diagnosis, and concurrent diagnosis including self-injurious behaviour and pica, namely the frequent eating or mouthing of things that are not food, which is a common eating disorder in children with autism. Other researches on ASD training programmes such as visual pedagogy – a non-traditional approach to behaviour guidance that takes advantage of the ability of children with autism to respond better to pictures rather than words – The Picture Exchange Communication System (PECS), Treatment and Education of Autistic and related Communication-handicapped Children (TEACCH), and sensory integrating programmes were able to decrease autistic mannerism and increase cooperation both in their own dental routine care and in the dental clinic setting [Limeres-Posse et al., 2014; Orellana et al., 2014]. Orellana and colleagues [2014] evaluated the compliance of ASD dental patients before and after a TEACCH-based training programme. They reported that after the programme, ASD patients were significantly more cooperative during dental treatment. Bäckman and Pilebro used a visual pedagogy to introduce to pre-school autistic children how to take care of their oral health and recommended dental treatment [Bäckman and Pilebro, 1999; Pilebro and Bäckman, 2005]. They found

that it significantly helped increase cooperation regarding their own dental routine care and in the dental clinic. In Thailand, there was a study using visual pedagogy to reduce negative behaviour and prepare autistic children for dental examination. This study showed that the training significantly helped in decreasing inappropriate behaviours such as self-injury and increased cooperation during dental examination [Ratanapiroj et al., 2007; Leklertsiriwong et al., 2007].

Although dental health and cooperation of ASD patients have been examined in a number of studies, there is limited knowledge in regards to Thai ASD patients. The aims of this study were to evaluate cooperation rates in Thai ASD patients and to analyse the factors associated with cooperation levels among ASD patients during dental treatment by means of cross-sectional analysis.

## Materials and methods

This study was approved by the Ethical Committee, Faculty of Dentistry and Faculty of Pharmacy, Mahidol University Institutional Review Board (MU-DT/PY-IRB 2015/048.3009) and Rajanukul Institute. Consent was received from all parents/legal guardians. All subjects were free to withdraw from the study at any time without affecting their dental treatment. Statistical consultation was done before sample size calculation based on the McNemar test using the nQuery Advisor programme. With type I error=5%, Type II error = 10%, power=80%, the minimum participant number was 88 enough for statistic achievement [Matshall et al., 2007]. The total number of subjects would be 106, to make up for any possible lost cases (20%).

### Subject selection

There were 103 subjects initially. Eight of them did not return the questionnaire so they were excluded, so the total number of participants was 95. Subjects were selected among children aged 3–18 years who were diagnosed with ASD. This was from the DSM-5 assigned by medical specialist, paediatrician or psychologist. The children came for dental treatment at the Rajanukul Institute, which is the governmental agency under the Department of Mental Health, Ministry of Public Health (MOPH), Bangkok, Thailand. This institute has a policy to extend medical and dental services to people in the community including special needs children and to provide knowledge, early intervention and programmes to improve child skills. All subjects selected to participate in this study were evaluated for sensory processing by an occupational therapist. Subjects whose parents or legal guardian desired to discontinue the participation in the study or did not return completed questionnaires were excluded.

### Study design

This is a cross-sectional study which is a type of

observational study that involves the analysis of data collection from selected participants [Matshall et al., 2007]. All the chosen subjects were receiving dental treatment in the regular dental clinic. Data was collected from patient's medical records and parental questionnaires as well as behaviour rating during dental treatment.

### Data from medical records

The medical charts of all subjects were reviewed and classified into seven categories: 1. Subject's demographic information (age and gender); 2. Concurrent diagnosis (ADHD, epilepsy, mental impairment); 3. Treatment aspects (psychiatric medication therapy, occupational therapy and behavioural modification, speech therapy); 4. Past dental history; 5. Previous history of dental or medical procedure under general anaesthesia; 6. Sensory integrating problems evaluated by an occupational therapist (auditory, visual, vestibular, proprioception, tactile, gustatory/olfactory systems); 7. Past dental procedures (oral examination/prophylaxis and fluoride application, operative treatment without the injection of local anaesthesia, operative or surgical treatment with the injection of local anaesthesia).

### The questionnaire

All participants' parents were asked to fill the questionnaire. All questions were close-end answers. Seven categories of questions were asked: 1. Medical and dental history; 2. The presence of aggressive behavior; 3. Level of receptive language according to ability to follow the instructions (cannot follow instructions, simple step instructions, multiple step instructions); 4. Level of expressive language (non-verbal communication, echolalia, moderate to normal ability to use the language); 5. School setting (regular class/integrated school, special education, none/home-school); 6. Oral hygiene care (full parental engagement, child's self-response, child's self-response with parental supervision, none); 7. Toilet trained (yes/no).

### Behaviour rating

The dental procedures received by the subjects on the day of data collection were categorised into three groups: 1. Oral examination and preventive procedures including oral examination, dental polishing and fluoride application. These procedures have few stimuli causing patients no or low pain; 2. Operative treatment without the injection of local anaesthesia, which has more stimuli such as sound and vibration from the high-speed and low-speed handpieces; and 3. Operative or surgical treatments with the injection of local anaesthesia, which include procedures such as filling in deep cavity, pulpotomy, pulpectomy, SSC or extraction. Patient's behaviour was rated by five dentists (kappa co-efficiency=0.75) experienced in treating ASD children using the Frankl scale, a reliable and frequently used behaviour rating systems in both clinical dentistry and research in ASD

Characteristics		N (%)
Gender	Male	82 (86%)
	Female	13 (14%)
Age (years)	3-7	33 (35%)
	8-11	43 (45%)
	12-18	19 (20%)
Concurrent diagnosis	None	65 (68%)
	ADHD	25 (26%)
	Epilepsy	5(5%)
Mental Impairment	Yes	81 (85%)
	No	14 (15%)
Aggressive behaviour	Yes	37 (39%)
	No	58 (61%)

TABLE 1 ASD subjects characteristics in this study.

patients [AAPD, 2013-14]. This scale separates observed behaviours into four categories ranging from definitely negative to definitely positive (Rating 1: Definitely negative; Refusal of treatment; crying forcefully, fearful, or any other evidence of extreme negativism; Rating 2: Negative. Reluctance to accept treatment; uncooperative; some evidence of negative attitude but not pronounced, i.e., sudden withdrawal; Rating 3: Acceptance of treatment. at times cautious: willingness to comply with the dentist, at times with reserve, but patient follows the dentist's directions cooperatively; Rating 4: Definitely positive. Good relationship with the dentist; interested in the dental procedures; laughing and enjoying the situation).

### Statistical analysis

Qualitative data from medical records, questionnaires as well as behaviour ratings were analysed using descriptive analysis. For factors associated with clinical behaviour during dental treatment, Chi-square test and multiple logistic regression analysis were used at the p-value < 0.05 level of significance. All statistical analysis was performed using PASW-18 software (Microsoft Corporation, CA, USA).

## Results

### Subjects' characteristics

Ninety-five ASD patients participated in the study (Table 1). Mean age was  $8.7 \pm 3.9$  years. The medical records indicate that some of the subjects were prescribed psychiatric drugs such as Methylphenidate or Risperidone. Eighty-nine per cent of subjects had received dental treatment in the past. Table 2 shows that most of the ASD patients had undergone behaviour modification treatment, such as speech and occupational therapies.

Most of the subjects had no sensory integrating problems. However, tactile was found to be highest among all sensory integrating problems listed. Most subjects were enrolled in normal or integrated schools and able to communicate using words or echolalia. Most of the parents were fully responsible for the child’s oral care. With regards to toilet training, most of subjects had been trained and able to use the toilet themselves.

**Behaviour rating**

Behaviour rating was divided into two groups using the

Frankl rating scale: uncooperative or negative behaviour, and cooperative or positive behaviour. It is found that before treatment 36% and 64% of the patients showed negative and positive behaviour, respectively. During the treatment procedures, the dentists determined that 55% and 45% of the patients exhibited an uncooperative and cooperative behaviour, respectively.

The behaviour was evaluated again after treatment. The score indicates that 27% of the patients showed negative behaviour, and 73% expressed a positive behaviour.

Medical and dental history		N (%)
Psychiatric medication used	Yes	32 (34%)
	No	63 (66%)
Previous dental treatment	Yes	85 (89%)
	No	10 (11%)
Previous general anaesthesia	Yes	27 (28%)
	No	68 (72%)
Previous hospital admissions	Yes	45 (47%)
	No	50 (53%)
Speech therapy	Yes	64 (37%)
	No	31 (33%)
Occupational therapy	Yes	75 (78%)
	No	20 (21%)
Past dental procedures	Preventive procedure	54 (57%)
	Operative treatments with the injection of local anaesthesia	18 (19%)
	Operative treatments without the injection of local anaesthesia	23 (24%)
<b>Sensory integrating problems</b>		
Auditory system	Yes	12 (13%)
	No	83 (81%)
Visual system	Yes	9 (9%)
	No	86 (91%)
Vestibular system	Yes	40 (42%)
	No	55 (58%)
Proprioception system	Yes	41 (43%)
	No	54 (56%)
Tactile system	Yes	57 (60%)
	No	38 (40%)
Gustatory / Olfactory system	Yes	5 (5%)
	No	90 (95%)
<b>Patients' skills</b>		
Education	Normal/Integrate	41 (43%)
	Special	38 (40%)
	None	16 (17%)
Verbal communication	No	30 (31%)
	Words/ Echolalia	35 (37%)
	Sentences	30 (32%)
Follow instruction	No	6 (6%)
	Simple	72 (76%)
	Complex	17 (18%)
Tooth brushing	No	1 (1%)
	By parent	38 (40%)
	By Children themselves	28 (29%)
	By both	28 (29%)
Toilet trained	No	21 (22%)
	Yes	74 (78%)

**TABLE 2** Data of all ASD subjects from medical and dental records and parental questionnaires.

Patient characteristics	Behaviour during dental treatment N (%)		P-value
	Uncooperative	Cooperative	
<b>Age</b>			
3-7	24 (72.73%)	9 (27.27%)	<.001*
7-11	25 (58.14%)	18 (41.86%)	
11-18	3 (15.79%)	16 (84.21%)	
<b>Behaviour before treatment</b>			
Uncooperative	31 (91.10%)	3 (8.80%)	<.001*
Cooperative	21 (34.40%)	40 (65.60%)	
<b>Dental procedures</b>			
Preventive procedures	20 (37.04%)	34 (62.96%)	<.001*
Operative treatment performed without local anaesthesia	19 (82.61%)	4 (17.39%)	
Operative or surgical treatment performed under local anaesthesia	13 (72.22%)	5 (27.78%)	
<b>Sensory integrating problems</b>			
<b>Proprioception system</b>			
No	24 (44.44%)	30 (55.56%)	.021*
Yes	28 (68.29%)	13 (31.71%)	
<b>Tactile system</b>			
No	16 (42.11%)	22 (57.89%)	.043*
Yes	36 (63.16%)	21 (36.84%)	
<b>Skills</b>			
<b>Education</b>			
Normal / Integrated	15 (36.59%)	26 (63.41%)	.004*
Special	24 (63.16%)	14 (36.84%)	
None	13 (81.25%)	3 (18.75%)	
<b>Verbal communication</b>			
No	23 (76.67%)	7 (23.33%)	.007*
Words / Echolalia	18 (51.43%)	17 (48.57%)	
Sentences	11 (36.67%)	19 (63.33%)	
<b>Follow instructions</b>			
No	3 (50.00%)	3 (50.00%)	.014*
Simple	45 (62.50%)	27 (37.50%)	
Complex	4 (23.53%)	13 (76.47%)	
<b>Tooth brushing</b>			
No	1 (100.00%)	-	.014*
By parent	28 (73.68%)	10 (26.32%)	
By child	12 (42.86%)	16 (57.14%)	
By both	11 (39.29%)	17 (60.71%)	
<b>Toilet trained</b>			
No	18 (85.71%)	3 (14.29%)	.001*
Yes	34 (45.95%)	40 (54.05%)	

**TABLE 3** Factors associated with cooperative behaviour of ASD subjects during dental treatment.

### Factors associated with patients' behaviour during dental treatment

Table 3 shows that the factors significantly associated with the behaviour during dental procedures are age, behaviour before dental treatment and dental procedures ( $p < 0.001$ ). Results also show that other factors such as gender, concurrent diagnosis including mental impairment and the presence of aggressive behaviour have no association with behaviour during dental treatment. The variables behavioural modification medicines, dental history, previous history of dental or medical procedures under general anaesthesia, speech and occupational therapies show no association with behaviour during dental treatment. Regarding six factors of sensory integrating problems which are auditory, visual, vestibular, proprioception, tactile, and gustatory/olfactory systems, only two of them are significantly associated with behaviour during dental treatment. These are the proprioception system ( $p = 0.021$ ) and the tactile system ( $p = 0.043$ ). While taking skills into consideration, all factors in this group are statistically associated with behaviour during dental treatments.

Multiple logistic regression analysis was performed to identify factors associated with patient's levels of cooperation during dental treatment. Results show that age, education, and behaviour before dental treatment are associated with the cooperative behaviour of ASD patients during dental treatment. The level of cooperation during dental procedures increased with the age of the patients. Table 4 shows that the level of cooperation among ASD dental patients aged 11–18 years is 11 times greater than patients aged 3–7 years (adjusted OR=11.51,  $p = 0.004$ ). Results also demonstrate that patients who had attended the special education system have a greater cooperation level than patients who did not, almost 13 times (adjusted OR=12.7,  $p = 0.014$ ). The analysis also found that patients who showed a positive behaviour before the dental procedure were more likely to show higher cooperation during the dental treatment, which was 15 times greater than in patients with negative behaviour before treatment (adjusted OR=15.26,  $p < 0.001$ ).

## Discussion

In this study, ASD has a higher incidence in males which is in agreement with previous studies [Kogan et al., 2009; Kim et al., 2011; Loo et al., 2008; Marshall et al., 2007]. For the cooperation level, subjects who showed positive behaviour before dental procedure were two times more likely to cooperate during dental treatment than those exhibiting negative behaviour before treatment. However, during dental treatment, the number of uncooperative patients increased to 55%, similarly to a previous study [Loo et al., 2009]. The level of cooperation is influenced by many factors such as patient characteristic, medical and dental history, sensory integrating problem, and patient life

Factors	P-value	Adjusted odd ratio (95% CI)
Age		
3–7	-	1
7–11	.138	2.36 (0.76, 7.31)
11–18	.004*	11.51 (2.22, 59.74)
Education		
Normal / Integrated	.153	4.33 (0.58, 32.27)
Special	.014*	12.70 (1.67, 96.98)
None	-	1
Behaviour before treatment		
Uncooperative	-	1
Cooperative	<.001*	15.26 (3.67, 63.32)
Multiple logistic regression analysis, * Significant difference at $P \leq 0.05$		

**TABLE 4** Multiple logistic regression analysis of factors associated with cooperative behaviour of ASD subjects during dental treatment.

skill [Marshall et al., 2007; Orellana et al., 2014]. The results of this study indicate that age, proprioception and tactile systems problems, behaviour before dental treatment, experience of behaviour shaping, verbal communication, ability to follow instructions, tooth brushing habit, and toilet training are associated with the cooperation level of ASD subjects during dental treatment. This is consistent with the study by Marshall and colleagues [2007], who found that patients older than 4 years, either non-verbal or displaying echolalia, unable to follow instructions, with parents carrying out tooth-brushing, and partially or not toilet trained (items related to the level of cooperation) generally have uncooperative behaviour during dental treatment [Marshall et al., 2007].

The relationship between tactile system problems and cooperation level during dental treatment are also similar to previous studies, which found that patients with enhanced sensitivities dislike the feeling of the toothbrush in their mouth resulting in excessive gagging or overreaction, or even as face and mouth were touched. Routine brushing can help familiarise the patients' with this feeling and reduce uncooperative behaviour during dental treatment. A firm instead of light touch be used [Stein et al., 2011; Stein et al., 2014].

This study identified three predictors of ASD patients' behaviour during dental treatment: age, experience of behaviour shaping, and behaviour before dental treatment, which is in line with the study of Loo and colleagues [2009]. This could be explained by the nature of ASD. Naturally, ASD is not a deteriorating disorder. Subjects can improve with time and learning. Considering age as a predictor, as the patients grow up their experiences lead to knowledge and cognitive development which influence behaviour. As for education, when ASD patients are educated in a special education setting, they are encouraging to develop physically, emotionally, mentally and intellectually through appropriate activities. The education content is

designed to promote nine important skills: intention, imitation, recognition, self-sufficient, sociability, fine motor development, physical movement, knowledge and communication skills. Rauf and colleagues [2014] found that most of the children with ASD who enrolled in special education school showed average adaptive behaviour (keep up with numbers and time), independent functioning, self-direction, physical development, responsibility, and socialisation. Taking positive behaviour before the dental treatment into consideration, the behaviour might be influenced by past experiences or by the patients' temperament. From a psychological standpoint, temperament can be defined as a natural difference in behavioural style causing individuals to react to stimuli variously. It was found that negative behaviours are frequently found in more reactive and less adaptive children. On the other hand, children with lower reactivity and higher adaptability seem able to cope with stressors and have a positive behaviour [Holmbeck et al., 2007].

The limitations of our study include its cross-sectional design, the possible inaccuracy of a parental report of ASD diagnosis, and dental care being provided by multiple clinicians. Due to time constraints, factors associated with patients' medical and dental history were taken from the medical records which contained the diagnosis but no record of severity. Severity of autism can also affect the level of cooperation. This study focuses only on whether or not the patients have experience in the dental clinic; however, the different frequency and experiences the patients have could result in the different level of cooperation.

## Conclusion

Based on this study, it can be concluded that school-aged ASD children who have been trained in special education programmes and showed positive behaviour before dental treatment exhibit a higher cooperation during dental treatment.

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