

Status Quo and Advanced Progress in Oral Health Care and Treatment of Children with Autism Spectrum Disorder: A Literature Review

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Autism spectrum disorder (ASD) has become one of the fastest growing diseases in the world, causing a great burden to ASD children's families and society. Children with ASD face more disadvantages relating to their oral health than those without ASD. There is a positive correlation between prevalence of caries lesions and severity of ASD. Poorer oral hygiene, higher detection rates of dental calculus and far more frequent cases of gingivitis occur in children with ASD. Traumatic injuries and various types of malocclusions are more frequent in children with ASD. Poorer oral health care and treatment status are caused by multiple adverse factors. Ways of promoting effective oral health care and treatment include pretreatment counseling; improvement of the individualised treatment environment; routine behaviour guidance techniques (BGTs) including tell-show-do, distraction, role model presentation, voice control, visual education and social stories, encouragement and reinforcement; targeted BGTs including visual education, behaviour modelling, applied behaviour analysis (ABA) and systematic desensitisation; passive BGTs including protective restraint, pharmaceutically administrated sedation and general anaesthesia; oral health education for guardians; and interdisciplinary collaboration and professional dental care/treatment. Dentists, families with children with ASD and schools should cooperate to improve family-centred oral health care and treatment for ASD children not only in China, but also the whole world.

Key words: autism spectrum disorder; behaviour management, children, oral health care, promotion

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Autism spectrum disorder (ASD) was first reported by Professor Kanner in 1943¹. Researchers from various countries studied similar cases and continuously adjusted the diagnostic boundary criteria of ASD spectrum^{2,3}. In the diagnostic and statistical manual of disorders⁴, ASD was defined as a single category, whereas subcategories such as Asperger's syndrome and pervasive devel-

opmental disorder not otherwise specified (PDDNOS) were removed. ASD has become one of the fastest growing disorders in the world. In 2011, the prevalence rate in the United Kingdom was 1.6%⁵, then in 2018, it was reported to have exceeded 1/50 in the United States⁶. In 2019, the prevalence rate exceeded 1/100 in China, and it continues to rise^{2,7,8-12}. The pathogenesis of ASD is still not entirely clear. It causes a great burden to families of children with ASD and society.

Children with ASD face greater disadvantages concerning their oral health than those without. Multiple adverse factors lead to poorer oral health care and treatment status, including perioral muscle characteristics and eating habits, perioral paraesthesia, oral microbial environment and drugs, bad oral habits, self-injurious behaviours, communication skills and behavioural characteristics, and unbalanced medical service resource allocation^{1,2,8,13,14}. Various generalised and specialised methods of effective oral health care in children with

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ASD have been explored and need to be optimised further in larger populations, such as pretreatment counselling, individualised treatment environments, routine behaviour guidance techniques (BGTs), targeted BGTs, passive BGTs, oral health education for guardians, interdisciplinary collaboration, and professional dental care/treatment¹⁵⁻¹⁹; however, the current situation and advanced progress on oral health care and treatment for children with ASD have not been updated, summarised or analysed.

This review sought to provide an overview of the current situation in oral health care and treatment of children with ASD, influencing factors of oral health status in children with ASD, and ways to promote effective treatment and daily oral health care in children with ASD.

Oral health status of children with ASD

Dental caries

There is a positive correlation between the prevalence of caries and the severity of ASD^{2,13}. Compared with children without ASD, those with ASD have a higher incidence of caries²⁰. A meta-analysis conducted by Da Silva et al²¹ showed that the caries prevalence rate in children with ASD is around 60.6%. The caries prevalence of children aged 3 to 5 years and with ASD is 50% to 60% in some areas of China, which is close to the national average (the fourth Chinese Oral Health Epidemiological Survey found that the prevalence of primary teeth in 3-, 4- and 5-year-old groups was 50.8%, 63.6% and 71.9%, respectively²²). Insufficient frequency of tooth brushing and frequent consumption of sugary foods and drinks contribute most to this^{2,8}.

Periodontal status

Children with ASD have poor oral hygiene. The detection rates of gingival bleeding and dental calculus are significantly higher than those for children without ASD¹⁴, and almost all children with ASD suffer from gingivitis^{22,23}. The latter are also more likely to have bad breath and food impaction^{8-10,12}. Insufficient daily oral health care, such as tooth brushing, and use of psychotropic drugs or antiseizure drugs (e.g., phenytoin sodium) are associated with the status^{2,8,24}.

Dental trauma

There is a significant difference in the prevalence of dental trauma between children with ASD and those with-

out. Marra et al²⁵ found that girls with ASD (57%) suffer more traumatic injuries than boys with ASD (43%). The comparison of different dental trauma types (enamel fracture, enamel/dentine/pulp fracture, root fracture, avulsions) and different soft tissue lip injury locations (upper and lower lip) for the ASD group and non-ASD group showed a statistically significant difference ($P < 0.05$)²⁵. The lower lip was found to be injured more often than the upper lip ($P < 0.005$). Furthermore, the long-term extraction rate of traumatic teeth in children with ASD is higher than that in those without ASD²⁵. Oral self-injurious behaviours in children with ASD often lead to traumatic ulcers and tooth loss²⁶. In addition, children with ASD are prone to tooth grinding, tongue thrusting and chewing/swallowing foreign bodies, resulting in an increased risk of damage to the oral soft and hard tissues^{27,28}.

Malocclusions

Self-injurious behaviours and unhealthy oral habits (bruxism, tongue thrusting, lip biting, thumb sucking and object chewing) contribute to the incidence of relevant malocclusions in children with ASD^{29,30}. Skeletal and dentoalveolar deformation may occur due to the frequency and severity of the behaviours and habits³¹. There is no specific type of malocclusion for children with ASD, though a tendency towards increased horizontal overlap, increased anterior open bite, increased posterior reverse articulation and a deeper palatal vault has been observed³¹⁻³³.

Influencing factors of oral health status in children with ASD

Influencing factors of oral health

Perioral muscle characteristics and eating habits

Dietary habits, oral hygiene habits and oral environmental factors of children with ASD affect their oral health care and promote caries lesions and gingivitis. Children with ASD have greater perioral muscular tension and poorer tongue flexibility, resulting in insufficient coordination of chewing and swallowing movements, which leads to food being contained in the mouth. They also prefer soft and sticky foods, which are likely to become attached to the teeth. In addition, desserts are often used as a reinforcement for behavioural rehabilitation training, leading to increased sugar consumption and greater risk of caries lesions^{1,2,8-10}.

Perioral paraesthesia

Perioral paraesthesia occurs in more than 90% of children with ASD and is mainly manifested as hypersensitivity. The physical stimulation of tooth brushing or taste stimulation of toothpaste can cause discomfort and activate the gag reflex, increasing the risk of toothpaste being swallowed by accident. Children with ASD with poorer hand mobility and comprehension skills find it difficult to complete daily tooth brushing^{2,8,9,12}.

Oral microbial environment and drugs

Studies have found that compared to children without ASD, the oral flora of children with ASD consist of more periodontal pathogens, leading to a stronger gingival immune inflammatory response. In addition, children with ASD children often also experience epilepsy, depression and other symptoms and thus require long-term use of psychotropic drugs that affect the amount of saliva produced and flow rate, impairing oral self-cleaning and increasing susceptibility to caries; however, more evidence is required to establish the effect of psychotropic drugs on caries risk³⁴⁻³⁶.

Bad oral habits

The higher prevalence of malocclusions in children with ASD is due to bad oral habits such as tongue thrusting and mouth breathing. The main types of malocclusion are maxillary transverse deficiency, open bite or deep vertical overlap, mandibular retrusion and dental crowding. These malocclusions affect children's facial aesthetics, as well as their physical and mental health development. In addition, low awareness of how to protect themselves leads to an increased risk of anterior dental trauma in children with ASD with protruded anterior teeth. Furthermore, malocclusion can be accompanied by airway obstruction and sleep apnoea symptoms, increasing the adverse effect on growth and development^{1,8,13,25}.

Self-injurious behaviours

Children with ASD, especially those with impaired linguistic ability, have a significantly higher incidence of self-injurious behaviours. This may be due to their inability to convey physical pain. More than 75% of the injury sites consist of the head and neck regions, and the behaviours are manifested as stabbing of the gingival soft tissues with the fingers or foreign bodies, lip and cheek biting, and even extraction of teeth by children themselves³⁷⁻³⁹.

Influencing factors of treatment

Communication skills and behavioural characteristics

Children with ASD have poor communication skills, and it is difficult for them to express their feelings and describe their oral symptoms in a timely manner. Guardians often focus on the children's behavioural correction and rehabilitation training while ignoring their oral health status and appropriate timing of treatment.

Behavioural characteristics of children with ASD are the main factors affecting the treatment of oral diseases. These children are highly sensitive to sound, light, smell, touch and unfamiliar environments. Their dental fear is more severe than that of children without ASD. They exhibit multiple self-stimulating behaviours, including twisting hands, flapping arms, crying and other repetitive movements, and trying to escape from the treatment environment. They may even damage objects or hurt others by scratching, biting, kicking or head hitting. If there is no effective way to comfort them, these children are prone to injuring themselves and others, and indeed compromising the diagnosis and treatment process^{13,14,39,40}.

Unbalanced medical service resources

In China, the number of professional paediatric dentists is inadequate, medical service resources are unbalanced^{2,9,10} and paediatric stomatologists often lack behaviour management training for children with ASD. Families of children with ASD devote most of their energy and financial resources to behavioural correction and rehabilitation training, meaning they have limited energy and financial resources to spend on oral health care and treatment. In addition, ignorance and implicit discrimination still exist towards children with ASD and their families. All these factors increase the difficulties medical treatment poses for children with ASD^{2,8-13}.

Promotion methods of effective treatment in children with ASD

Pretreatment counselling

Prior to treatment, guardians of children with ASD should communicate with stomatologists to prepare for the treatment process and be able to cooperate with doctors about the necessary behaviour management. Guardians should provide information about their children's unique preferences and specific behaviours; this is of great value for doctors in developing personalised be-

behaviour guidance methods. Professionals should learn about the children's previous diagnoses and treatment history for oral diseases and concomitant diseases, developmental age and status, life skills, language expression level and reading ability^{2,41}.

Treatment environment

The treatment environment should be changed based on the needs of children with ASD to relieve anxiety and reduce resistance behaviours so they can gradually become familiar with their new environment and adapt to it. An independent waiting area and single treatment room should be reserved to ensure the same environment for each visit. Relaxing and slow music or white noise can be added, and lighting conditions should be improved^{2,42}.

Routine BGTs

BGTs are crucial to ensure a smooth treatment process and are used widely in paediatric dental treatment, including tell-show-do (TSD), distraction, role model presentation, voice control, encouragement and reinforcement^{17,18,43-45}. Children with ASD often experience attention and comprehension deficit; thus, the effect of single BGT methods is limited, and it is often necessary to combine multiple methods in real situations^{19,45}.

Encouragement and reinforcement is an important and necessary BGT (especially for children aged from 3 to 6 years) to improve cooperation of in children with ASD⁴⁶. Forms of reinforcement include doctors' pronunciation and intonation, language content, facial expressions, body movements and material objects. For children with ASD with understanding and attention deficit, simple linguistic encouragement (e.g. "you're great!") is not sufficient. Doctors can choose tangible rewards based on children's preferences. The frequency and timing of rewarding also influence the reinforcement effect. Too high a frequency will reduce the effect, whereas too low a frequency will fail to counter the effect of negative stimuli. Specific methods should be used according to the specific situation⁴⁵.

Targeted BGTs

Targeted BGTs can be used to improve the quality and efficiency of treatment. Individualised BGTs should be adopted based on the specific situation, and a combination of techniques can be applied. Persistence of guardians and oral health professionals is key.

Visual education

Children with ASD are often visual thinkers. As such, visual education methods are effective for them. By conveying information through video and images, visual education helps children with ASD to learn relevant skills⁴⁵. They and their guardians can learn from pictures and videos of dental clinical processes to repeatedly exchange information with the outside world and become familiar with the treatment environment¹⁹.

Narzisi et al⁴⁷ used MyDentist software (Bilbao, Spain) to provide rich and personalised image and video material for children with ASD. With its social story models and relaxing games relating to oral disease treatment, this software helps children with ASD become familiar with the treatment process⁴⁷.

In addition, with the development of information technology, the application of virtual reality (VR) technology is increasing; however, VR technology is not used widely in the treatment of oral diseases. It serves primarily to create a safe and progressive virtual environment using music, videos and game scenes so as to distract attention, make ASD children more relaxed and better adapt to the treatment environment. At present, the equipment requirement threshold and technology sensitivity of VR limit its wide application. In addition, its ethical significance should be considered fully¹⁸.

Behaviour modelling

Behaviour modelling is based on the rigid repetitive behaviour characteristics of children with ASD. By using certain ingrained behaviours or habits, this method can help control the dietary habits and daily oral health behaviours of children with ASD. The effect of this method largely depends on long-term training implanted by guardians; however, once a good behaviour pattern is formed, daily oral health care will be simplified greatly⁴⁵.

Applied behaviour analysis

Applied behaviour analysis (ABA) is a successful psychological treatment approach for patients with ASD and has been validated for its effect of improving various behaviours. By dividing complicated target tasks into simpler ones and training patients with ASD step by step using the appropriate reinforcements, this method helps such patients finally complete complicated target tasks. ABA has been used successfully to improve the performance of children with ASD when sitting still and during intravenous injection, physical examination, oral

examination, pit and fissure sealing and periapical radiograph examination⁴⁶.

The following steps can be used for ABA training: repeatedly shining a light into children's mouth with a flashlight or mobile phone torch and helping children learn to open and close their mouth appropriately using demonstration if necessary; gradually illuminating children's teeth with oral mirrors and counting their teeth with probes; and helping children to sit in dental chairs, encourage them to contact treatment tools. Attention should be paid to any changes in children's facial expression and reinforcers should be used in a timely manner¹⁹. It should be noted that when unexpected behaviours occur, the best approach is to ignore them and wait for children to stop these behaviours^{19,48}.

Systematic desensitisation

The basic principle of systematic desensitisation is reciprocal inhibition, helping patients weaken and eventually cut the connection between stimuli and their anxiety responses⁴⁹. Step-by-step clinical activities help children with ASD to overcome their fear of the clinical environment and treatment process. Systematic desensitisation can also be combined with other BGT methods.

Researchers at the Affiliated Stomatology Hospital of Tongji University used systematic desensitisation and found it to have an obvious effect on the treatment of children with ASD⁴⁵. Most of the children trained made significant improvements in their cooperation⁴⁵. Xia⁵⁰ focused on oral tactile disorders in children with ASD (sensory input disorders relating to the oral cavity, vestibule and skin), helping them to reduce oral sensory sensitivity and improve oral sensation through systematic desensitisation.

Passive behaviour control techniques

Protective restraint

When facing emergencies or prior to procedural sedation, children with ASD may need to be placed in protective restraints to prevent uncontrolled body movements that could endanger their own safety and that of others.

Use of protective restraints for children with ASD is now controversial. A study showed that pressure on the body can help children with ASD remain emotionally stable (deep pressure theory), and appropriate physical pressure can provide a calm and comforting effect⁵¹. Medical professionals should take the time to explain the method and process to guardians and receive informed consent prior to using protective restraints.

During the treatment process, close attention should be paid to the tightness of restraints to avoid unforeseeable damage due to movements children may make while struggling². For children with ASD who are older and stronger, it is difficult to use protective restraints^{52,53}.

Pharmaceutical administered sedation

Comfortable treatment techniques including local anaesthesia, nitrous gas sedation and treatment of oral disease under general anaesthesia are a developing trend in dental treatment, especially in paediatric dentistry, providing new opportunities for children with difficulties to cooperate with routine treatment procedures^{2,45}.

Nitrous oxide (NO) sedation is the most convenient sedation method that can be controlled by dentists with a fast effect, rapid metabolism and no burden on the child's body⁵⁴. A study showed that the success rate of treatment with 50% NO in children with ASD reaches 87.5%⁵⁵. However, other studies have shown that children with ASD with 5,10-methylenetetra hydrofolate reductase (MTHFR) gene mutation suffer from folic acid metabolism disorder, and thus can die from long-term exposure to high-concentration NO under general anaesthesia, indicating that MTHFR deficiency is a contraindication for NO sedation^{56,57}. Currently, the feasibility or effect of NO sedation in oral disease treatment of children with ASD has not been validated fully, but there is no evidence of it having a fatal impact.

It has been reported that the combination of oral benzodiazepines (e.g., diazepam and midazolam) and NO sedation has a success rate of 77% to 100%^{51,58,59}. Careful planning and evaluation should be carried out by qualified physicians⁵⁹. Doctors should discuss the benefits and risks of NO sedation with guardians before making decisions^{2,45}.

General anaesthesia

ASD is one of the most common indications for oral disease treatment under general anaesthesia. When children with ASD cannot cooperate with sedation, general anaesthesia can help reduce treatment time and relieve the psychological burden on guardians⁶⁰. Informed consent must be obtained and indications and contraindications must be controlled strictly⁶⁰. Adverse events related to general anaesthesia in children with ASD include disruptive behaviour, postoperative vomiting, postoperative bleeding and postoperative seizures. The latter two are relatively rare⁶¹. Dentists should explain the ASD diagnosis, its complications and any potentially risky preoperative behaviours to anaesthesiologists to make a

perioperative presurgical plan⁶². Guardians and medical professionals should help desensitise children with ASD so they can adapt to the treatment process and hospital environment in advance^{2,45}.

Methods for promoting daily oral health care in ASD children

Visual education and social stories

Visual education and social stories have been used widely to help children with ASD learn various skills. Visual education has been described in the previous section.

Social stories are typically short stories that describe common social experiences and clarify what is happening in a particular situation and what the appropriate behaviours are^{63,64}. They play an effective role in teaching children with ASD oral health care skills^{65,66}. Du et al¹⁵ combined visual education and social stories to help children with ASD learn how to master tooth brushing skills. The 119 preschool ASD children they studied showed significantly improved tooth brushing skills and oral health status after 3 to 6 months of training^{15,44}.

Visual education in various forms (pictures, video, software, etc.) can help preschool children and adolescents with ASD improve their oral health care skills. Effective observation and continuous training are critical to children's performance^{43,44,67,68}. Children with ASD of higher intelligence will benefit more from the social story method because social stories require comprehension skills^{64,65}.

ABA

When faced with resistance from children with ASD, daily tooth brushing can be split into the following smaller tasks: massage and touch the child's head, neck, face, nose and lip; gently sweep the child's hard palate, buccal mucosa, gingiva and tongue with the toothbrush; and add water to the toothpaste, and gradually transfer to tooth brushing^{19,48}.

Oral health education for guardians

Guardians play a central role in the long-term oral health care of children with ASD. Oral health education for guardians can compensate for the information imbalance between families and oral professionals and help guardians adopt individualised methods to better deal with possible emergencies⁶⁹⁻⁷¹.

To overcome multiple difficulties, guardians explore a variety of methods with great patience, love and creativity, including:

- Making plans: ritualising the tooth brushing process and recording it on a calendar as a reminder, and providing a clear time frame by counting, singing or using a timer¹⁶;
- Acting as a role model: the whole family complete oral health care at the same time or demonstrate for children with ASD, and use visual education materials¹⁶;
- Incorporating amusement: creating a game situation when completing oral health care, and rewarding children with individualised reinforcers after oral health care¹⁶.

In addition, ASD children may swallow toothpaste which can lead to risks. Thus, fluoride toothpaste with a tolerable taste and texture should be tried and selected. Fluoridated milk, mouthrinse and other fluoride oral care methods can also be considered².

Interdisciplinary collaboration and professional level improvement

Interdisciplinary cooperation will play an effective role in improving children with ASD's oral health care status. If children have fixed psychiatrists and rehabilitation therapists, oral professionals should discuss with them to evaluate the children's characteristics, habits and skill learning status in order to offer personalised treatment^{66,72-74}. Although this method is time-consuming, as the number of psychiatrists and rehabilitation therapists grows, interdisciplinary collaboration will become more popular. The families of children with ASD hope for and require professional oral health care education and treatment^{75,76}; thus, oral health care professionals around the world should work harder to boost further cooperation.

The promotion methods for effective treatment and daily oral health care for children with ASD have been developed by clinicians and parents in the last few decades, especially in more developed cities such as Shanghai and Hong Kong^{15,44,45}. Nevertheless, the general status for children with ASD in China is still behind that of those in developed countries with more experience and greater exploration of different methods.

Further improvements should focus on family-centred oral health care education. Dentists, families of children with ASD and schools should cooperate more effectively to improve oral health care and help children with ASD of appropriate age receive timely oral

health care management, such as caries risk assessment, pit and fissure sealing and fluoride application. Furthermore, doctors' professional skills in treating children with ASD should be improved. Training in generalised and specialised BGTs and comfortable treatment techniques should be strengthened^{2,8}.

Conclusion

There is a positive correlation between prevalence of caries lesions and severity of ASD. Traumatic injuries and long-term extraction of traumatic teeth are more frequent in children with ASD, and various adverse factors cause them to have poorer oral health care and a poorer treatment status. Various generalised and specialised oral health care methods have been explored for children with ASD and need to be optimised further in the wider population. Dentists, families of children with ASD and schools should cooperate to enhance family-centred oral health care and oral health treatment services for children with ASD not only in China, but also in the whole world.

Conflicts of interest

The authors declare no conflicts of interest related to this study.

Author contribution

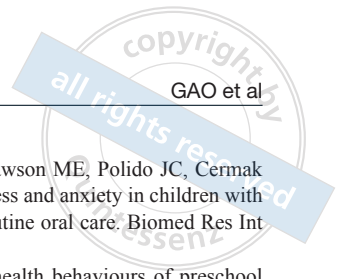
Dr Lu GAO contributed to the literature collection and drafting the manuscript; Dr Xue Nan LIU supervised the study design and reviewed and revised the manuscript.

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