#### **Case Report**

# Comprehensive Management of an Autistic Patient with Mental Retardation under General Anesthesia

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### Abstract

**Background:** Autism is a developmental disorder characterized by severe deficits in social interaction and communication and one of the most heritable complex genetic disorders in psychiatry. Comprehensive management of these children requires multidisciplinary teamwork. **Case Report:** A 16-year-old male patient was brought to the Department of Pedodontics and preventive dentistry by his parents with complaints of pain and sensitivity of teeth and consequent difficulty in eating in the lower left back tooth region for 2 weeks. Since the patient was combative in nature comprehensive management was planned under general anesthesia. Complete rehabilitation including pulpectomy, restoration, fluoride varnish application, and parental counseling was done. After the procedure, the patient recovered well from the pain discomfort he was having and significant improvement in his oral hygiene was noted. After 4 weeks, follow visit revealed significant improvement in his behavior.

Keywords: Autistic disorder, dental care for children, dental care for people with disabilities, epilepsy, oral health, special-care dentistry

## **CASE REPORT**

Autism is a developmental disorder characterized by severe deficits in social interaction and communication and one of the most heritable complex genetic disorders in psychiatry. Despite this high heritability, autism has a heterogeneous etiologic, with multiple genes and chromosomal regions likely to be involved. While no specific gene variant has been identified and confirmed that contributes to the expression of autism, it is very likely that several will be confirmed over the next decade. Twin and sibling studies demonstrate heritability in autism. A wide spectrum of medical and behavioral symptoms is exhibited by children with autism, which makes routine dental care very difficult in them. Autism is a neurobehavioral and cognitive disorder characterized by impaired development of interpersonal and communication skills, limited interests, and repetitive behaviors. The prevalence of Autistic spectrum disorders (ASD) is estimated to be 1% worldwide.<sup>[1]</sup> The incidence of autism is about 0.2%.<sup>[2]</sup> This complex mental disability is about four times more prevalent in males but is more severe in females. It manifests during the first 3 years of life.<sup>[3,4]</sup> It is important to recognize that for almost all cases, genetic testing is unlikely to establish a diagnosis of autism in the absence of careful clinical evaluation, since Fragile X

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Mental Retardation-1 gene, maternally inherited 15q11-q13 duplications, and other syndromes greatly increase the risk for autism, but do not lead to autism spectrum disorders in all cases. Epilepsy is an enduring predisposition for generating seizures which is active in approximately 0.4%-0.8% of the population. The majority of studies that examined sex differences in people with ASD found an increased risk in males, which is consistent with higher male prevalence for ASD in the general population. These results support the strong connection between intellectual disability, epilepsy, and ASD. Clarke et al. found that the greatest risk for developing ASD was in patients with seizure onset before 2 years. One further study found that 80% of children with both ASD and epilepsy had their seizure onset in the 1st year of life. This may be caused by the contributing role of disruptive seizures in ASD during early development<sup>[5]</sup> ASD and epilepsies are heterogeneous disorders that have diverse etiologist and pathophysiologies.

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The high rate of co-occurrence of these disorders suggests potentially shared underlying mechanisms. The systematic review done by Lauren found an overall pooled prevalence of 6.3% for ASD in epilepsy. Higher prevalence was found in studies with younger age, greater inclusion of intellectual disability, and specific epilepsy syndromes (infantile spasms and Dravet syndrome).<sup>[6]</sup> A systematic review by Strasser et al. found out that the prevalence of dental caries and periodontal disease in children and young adults with ASD can be considered as high, pointing to the need for oral health policies focused on these individuals.<sup>[7]</sup> Autistic children have significantly poor oral hygiene and a higher incidence of malocclusion and dental caries when compared to other oral conditions.<sup>[8]</sup> The behavioral symptoms in children include temper tantrums, hyperactivity, short attention span, impulsivity, agitation, anger, and a tendency for aggressive and self-injurious behaviors.[3] Disorders of language and social communication, poor response to external stimulation, tendency to isolate themselves, and poor eye-to-eye contact are well-recognized symptoms. Children with autism also commonly have damaging oral habits such as bruxism, tongue thrusting, picking at the gingiva, lip biting, and pica. This case report follows the care guidelines 2013 for reporting of case reports.

#### **Patient information**

A 16-year-old male patient was brought to the Department of pedodontics and preventive dentistry by his parents with complaints of pain and sensitivity of teeth and consequent difficulty in eating in the lower left back tooth region for 2 weeks. The mother reported that, during the previous week, her son had been shouting continuously, while pointing to the mandibular teeth on the left-hand side of his jaw. The parent noticed the pain was insidious in nature, which aggravated 2 days back on consumption of food. The boy had been undergoing routine therapy with a psychiatrist as well as medical rehabilitation in an attempt to train his motorics. However, he had never consulted a dentist. Significantly, the mother acknowledged that her son had been diagnosed with autism when he was 4-year-old and had recently attended a school providing additional teacher support. Based on the results of observations made during the first visit, the boy was known to be able to mimic words spoken by another person, although with imperfect articulation, responding to a conversation with a nod or a shake of the head. Unfortunately, two-way communication proved impossible since, during the consultation at the clinic, the boy was unable to calm himself, his eyes perpetually moving, while he continually produced incomprehensible language amounting to little more than sounds. The patient's medical history included a diagnosis of autism and epileptic not under medication. The patient had an episode of seizure attack 6 months back and he is now asymptomatic. The patient was aggressive and uncooperative for routine dental procedures so a complete rehabilitation was planned under general anesthesia. The family history was noncontributory. He had no history of drug allergies. The parents reported that the boy had a history of seizure for 3 years and was on medication phenytoin and stopped the medication for 3 months. The patient has gone to a nearby hospital with the same complaint, but no procedure was done as the patient was combative and uncooperative.

#### **Clinical findings**

The general examination revealed the patient was well built and nourished, height 160 cm, weight 94 kg, blood pressure 100/70 mmHg, PR 84 bpm at the time of admission. Routine blood investigations, thyroid-stimulating hormone, T3, T4, HIV test, Hepatitis B surface antigen, Hepatitis C virus, random blood glucose, S Calcium, blood urea, blood urea nitrogen, S Creatinine, serum electrolytes, liver functional test, and echography were done on the admission of the patient for the procedure under general anesthesia and the findings of the results were as follows [Tables 1-4].

#### **Timeline**

During the first visit, an extraoral examination was performed, the results of which revealed no facial asymmetry. From the outset of the consultation, the patient would sit on the dental chair, although only briefly, but did not want to open his mouth. The patient simply pointed with a finger to his left cheek. Oral hygiene instructions were emphasized during the first visit. The patient's mother was also counseled to train the child at home before the following visit, while the patient himself was asked to practice opening his mouth for 10 s before brushing his teeth every day. The second visit took place 1 month later. The mother complained that the child was experiencing pain in his left rear mandibular teeth. Unfortunately, the response of the child himself was consistent with that of his previous consultation. While the child remained agitated, tending to make sudden movements, he opened his mouth, even if only briefly, to be inspected. The examination was, therefore, limited to one using a dental mouth mirror. The results of the initial examination indicated the widespread presence of caries in the occlusal portion of tooth 36, retained root stumps 55, the occlusal caries of teeth 44, 45, and 46 as well as general plaque without staining. However, a radiographic examination could not be performed. Several treatment plans, including oral prophylaxis, tooth extractions, tooth restorations, and endodontic procedures were to be carried out under general anesthetic by multidisciplinary team of a pediatric dentist, pediatrician, anesthetist, and oral and maxillofacial surgeon.

#### **Diagnostic assessment**

The patient's mother was asked to help the dentist train the child to open his mouth by pretending to be a dentist herself. The patient's appointment was then rescheduled. However, obstacles occurred during the oral examinations of both children. For instance, the results of a provisional clinical examination indicated that invasive action in relation to certain teeth was required. However, the patients proved uncooperative and it was thus necessary to administer a pretreatment general anesthetic. According to certain literature, the behavior of ASD children in the clinic can actually determine appropriate

approaches. A number of experts also argue that a restraining approach may prevent the possibility of sudden aggressive behavior. The poor cognitive and motor abilities of the autistic patient prevented us from managing him on a dental chair. Since the patient was combative in nature and uncooperative for routine dental procedures a complete oral rehabilitation was planned under general anesthesia. The child was given fitness for the procedure by anesthetist, cardiologist, and endocrinologist. After getting informed consent from the parents and from the concerned departments the dental procedure was planned by pediatric dentist under general anesthesia.

The administering of nitrous oxide to ASD patients represents a challenge, given the prerequisite level of communication. Therefore, if the patient was unable to respond to a form of sedation using nitrous oxide, dental treatment involving the administering of a general anesthetic should be undertaken. Moreover, the need for extensive treatment (involving four quadrants) and/or complex treatment also triggers the use of general anesthesia.<sup>[9]</sup>

Patients with ASD will be very easily distracted. Consequently, certain steps, such as making physical contact with them during dental treatment, should be avoided. In addition, sensory stimuli (sound, odor, etc.,) should also be reduced in intensity.<sup>[9]</sup> Thus, before the initial examination of the patient, the examination chair had been adjusted to minimize potential distractions. Sudden movements of the chair in question were also avoided, as was the use of dental handpiece swivel instruments during the initial visit. Unfortunately, the intraoral examination was neither completely nor successfully performed during the first visit. Individuals with ASD generally experience hypersensitivity in the intraoral and perioral areas and tend to be sensitive to even a light touch during dental examination. Thus, refusal to cooperate or physical resistance during a dental examination is a distinct possibility with young ASD patients.<sup>[1,10]</sup> Similarly, previous research has already shown that most ASD children will demonstrate unhelpful behavior during dental examination.[11,12] Therefore, it is advisable that such patients be introduced previsit to clinical situations through pictorial stories or home exercises to familiarize them with tools and procedures, including the type of orders issued by dentists.<sup>[1]</sup>

#### **Therapeutic intervention**

During the third consultation, a full examination was performed after the patient had been placed under anesthesia in the operating theatre.

The patient was kept for nil per oral for 12 h before the procedure under general anesthesia, and the procedure lasted for about 2 h and the patient was shifted to postoperative intensive care unit and was monitored for 8 h and was shifted to postoperative ward.

The examination led to a diagnosis of pulpy-involved caries on tooth 46 with widespread clinical conditions in the occlusal and proximal mesials as well as enamel caries on teeth 16, 26, 34, 37, 44, and 45 [Figure 1].

The patient was prepped for the procedure. During the procedure, he was aggressive and combative with the support staff to manage for the induction of anesthesia as the patient kept touching the IV set on his hand due to his continuous movements of the hand. Subject was stabilized with extra bandage for protection. Several treatments were performed, namely: oral prophylaxis, dental restorative restorations on teeth 16, 26, 34, 37, 44, and 45 as well as fluoride application. After surgery, several instructions were issued to maintain oral hygiene, including: brushing of the teeth, compressing the wound area with gauze soaked in chlorhexidine, and following a soft food diet for 2 days. Management of postaction pain and fluid therapy was performed and monitored with a 7-h postoperative control subsequently being conducted. The patient was able to eat 15 h after surgery.

The patient's mother was asked to irrigate the operating area with chlorhexidine mouthwash and maintain the oral hygiene of the child by regular brushing. She also received an explanation about the prognosis of care. The patient then returned for a further consultation 3 weeks later during which he made no complaint of pain. During the fourth visit, the patient's behavior was more positive than during the first. For example, he proved able to briefly sit alone in the dentist's chair without being forced to. Moreover, the father confirmed that his son had started to brush his teeth regularly. The patient was then referred to the previous dentist for regular check-ups.

#### Follow-up and outcome

The parents were educated on the proper oral hygiene measures to be adopted and the need for regular dental visits in future. The patient was reviewed after 3 months and, again, after 12 months, by which time the clenching and grinding behavior had decreased significantly. His eating habits had also improved to a great extent because of the relief of the pain and discomfort. Keeping in mind the importance of prevention, a vigorous approach to preventive measures such as oral hygiene practices, dietary advice, and fluoride supplements were instructed. As a pediatric dentists responsibility was taken and services were offered in the prevention of dental disease in children with special health care needs by establishing communication with the authorities responsible for the welfare of these individuals and involving themselves in the total health care. Brushing with a fluoridated dentifrice twice daily was emphasized to help prevent caries and gingivitis. A noncariogenic diet was discussed for long-term prevention and if a diet rich in carbohydrates was



Figure 1: Access opening done with respect to 36

Table 1	l· C	omnlete	Metabolic	Panel
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Name of test	Test result	Average range
RBS (GOD-PAP method) (mg/dl)	100	70-140
Serum calcium (arsenazo method) (mg/dl)	10.9	8.8-10.2
Renal profile (mg/dl)		
Blood urea (modified berthelot)	21	10-50
BUN	9.8	4.7-23.3
Serum creatinine (Jaffe's alkaline picrate method)	0.8	0.7-1.4
Serum electrolytes (mEq/L)		
Sodium	146	136-145
Magnesium	5.1	3.5-5.1
Chloride	102	98-107
Liver function test		
Total bilirubin (mg/dl)	0.4	0.2-1.2
Direct bilirubin (mg/dl)	0.1	0.1-0.4
Indirect bilirubin (mg/dl)	0.3	0.2-0.8
Total protein (mg/dl)	6.8	6.2-8
Albumin (mg/dl)	4.4	3.5-5.5
Globulin (mg/dl)	2.4	1.8-3.5
A/G ratio (mg/dl)	1.83	1.2-2.2
Aspartate transaminase (SGOT) (U/L)	33	6-46
Alanine transaminase (SGPT) (U/L)	35	8-49
ALP (U/L)	177	88-306

ALP: Alkaline phosphatase, SGOT: Serum glutamic oxaloacetic transaminase, SGPT: Serum glutamic pyruvic transaminase, BUN: Blood urea nitrogen, GOD-POD: Glucose oxidase and peroxidase

Table 2: Serology Test		
Test done	Test result	
Reactive/nonreactive antibodies for HIV	Negative	
HBsAg	Negative	
HCV	Negative	
Reactive/nonreactive antibodies for HIV	Negative	

HBsAg: Hepatitis B surface antigen, HCV: Hepatitis C virus

medically necessary, the parent was asked to alter frequency besides increasing preventive measures.

The treatment greatly improved his eating habits as he could now chew solid foods. Long-term care included increasing the frequency and efficiency of oral hygiene measures with the help of the parents, application of fluoride gel or rinse, intake of healthy noncariogenic foods, and frequent recall appointments.

### DISCUSSION

Autism is an incapacitating disturbance of mental and emotional development characterized by severe deficits in social interaction and communication and the presence of repetitive, ritualistic behaviors. There are no specific genetic, medical, or laboratory tests available to confirm the diagnosis of autism and the comprehensive management of autism includes parental counseling, special education in a highly structured environment, speech therapy, and social skills training, with the ultimate goal of facilitating independence in activities of daily living

Table 3: Complete Blood Count		
Name of test	Test result	Average range
Blood grouping		
Blood group	А	
Rh typing	Positive	
CBC		
Hemoglobin (g %)	15.0	13.5-17.5
Total count (cells/cumm)	12,010	4000-11,000
Polymorph (%)	45.6	50-70
Lymphocytes (%)	39.7	20-40
Monocytes (%)	11.6	2-10
Eosinophil (%)	3.1	0.5-5
Platelet (Lakhs/cumm)	2.94	0.5-4.5
ESR (mm/h)	8	0-15
RBC (million/cumm)	5.28	3.5-4.5
PCV (%)	44.6	37-53
MCV (fL)	84.5	80-100
MCH (pg)	28.4	27-34
MCHC (g/dl)	33.6	32-36
RDW-SD (%)	13.5	11-16
PDW (fL)	42.4	32-56
PCT (%)	0.244	0.1-0.28
aPTT (s)	29.0	22-35
PT (s)	11.5	11-15
INR (s)	0.95q	PT control: 12.2

CBC: Complete blood count, RBC: Red blood cell count, MCV: Mean corpuscular volume, RBC: Red blood cell, MCH: Mean corpuscular hemoglobin, MCHC: Mean corpuscular hemoglobin concentration PT: Prothrombin time, PCT: Plateletcrit, aPTT: Activated partial thromboplastin time, INR: International normalized ratio, RDW-SD: Red cell distribution width – Standard deviation test, PDW: Platelet distribution width, ESR: Erythrocyte sedimentation rate

Table 4: Thyroid Function Test		
Test done	Test result	Average range
TSH (µg/ml)	0.05	0.25-5
T3 (ng/ml)	2.02	1.04
T4 (µg/ml)	10.81	4.2-11.8

and self-care. Children with autism have multiple medical and behavioral problems, which make their dental treatment extremely difficult. Communication problems and poor mental capabilities are central concerns when treating children with autism; these children exhibit wide variations in their ability to cooperate during dental treatment. For our patient, full-mouth rehabilitation was planned and was executed under general anesthesia because he suffered from severe mental retardation. Children with autism who have mild to moderate mental retardation and an absence of severe behavioral problems can be treated successfully in the general practice setting. Nevertheless, behavioral problems like hyperactivity and quick frustration can hamper the provision of oral health care in patients with autism. Furthermore, the invasive nature of oral care may trigger violent and self-injurious behavior such as temper tantrums or headbanging.<sup>[3]</sup> In our patient, it was very obvious that it would be difficult for him to comprehend instructions and cooperate on a dental chair, and hence the treatment was done under general anesthesia. General anesthesia gives the dental surgeon an opportunity to perform comprehensive and unhurried management of all problems in a single appointment.<sup>[13]</sup> Our patient was totally caries-free; this could be attributed to a spaced dentition, absence of any retentive area (flat occlusal surface due to bruxism), and open proximal contacts. Furthermore, the mother was well trained in the maintenance of the child's oral hygiene. In general, children with autism prefer soft and sweetened foods and they tend to pouch food inside the mouth instead of swallowing it due to poor tongue coordination, thereby increasing the susceptibility to caries.<sup>[14]</sup> Moreover, the risk for dental caries can be expected to be higher in these patients due to difficulties in brushing and flossing in them. Noninstitutionalized children with autism had caries rates that were similar to the rates in functionally independent peers in a study conducted by Klein et al. [15] Shapira et al. report that a combined treatment, provided by a dental team and a geropsychiatric team working together, resulted in a decreased prevalence of caries in a group of children with autism as compared to another group who did not receive any treatment.<sup>[16]</sup> Our patient presented with decayed teeth, which was corrected with stainless steel crowns. The option of using an intraoral appliance was not considered because it was unlikely that the child would comprehend and cooperate with this form of treatment and also because of the risk of aspiration. Rajic et al. reported a reduction in the frequency and severity of bruxism in an autistic child following injections of botulinum toxin into the masseter muscle.<sup>[17]</sup> Despite the good results and minimal side effects observed with this procedure, we did not opt for it because of its short duration of action, the lack of adequate information on the most effective dose and the best site of delivery of the injection, and the relatively high cost of treatment.<sup>[18]</sup> In addition, we were reluctant to carry out a procedure that is not commonly practiced in our region and we had doubts about the ability of our patient to tolerate it. The patient underwent extraction of the maxillary right primary central incisor; this tooth was nonvital and had an abscess associated with it. We did not attempt to preserve the tooth as there was only 1-2 mm of tooth structure above the gingival margin. We emphasized the need for routine dental check-ups and regular practice of oral hygiene measures to the parents. At the follow-up at 3 months and after 12 months, the mother reported a significant decrease in his pain and discomfort and clenching and grinding behavior. This might be attributed to the reduction in sensitivity to thermal stimuli following the placement of the stainless steel crowns.[19]

American Academy of Paediatric Dentistry's guidelines on Caries Risk Assessment has categorized them under High risk for physicians and other nondental health care providers and Moderate risk for dental providers. Attitude and knowledge of the oral health care professionals are of utmost importance while rendering oral health care to such children. The treatment rendered currently provides a long-term benefit for the patient. More focus had to be given for the long-term maintenance of oral health in special children. In addition, previous research suggests that dentists should educate parents of ASD children receiving dental care under general anesthesia about the risk of restoration failure. Actually, while there is no limit to the frequency with which an individual can be placed under general anesthetic, it is advisable to reduce the risk of anesthesia by minimizing the need for revisits. 3 As a result, restoration given to patients undergoing general anesthesia should be considered for its prognosis.<sup>[19]</sup> Another point to consider in planning dental care for a patient with impaired growth and development is the assessment of that individual's understands of the role and importance of oral care. Therefore, aggressive patients who refuse caregivers' assistance in maintaining oral hygiene will not achieve promising restoration results. Similarly, patients who are physically incapable of brushing or flossing cannot achieve any complex dental restoration since cooperation and careful attention play an important role in its success. Finally, it can be concluded that dental care for children with ASD should take account of the safety benefits and risks to both patient and dental teams. The dental care provided to children with ASD should also be supported by preventive efforts on the part of parents/caregivers and children. Moreover, general anesthesia may be considered to be a valid solution if other behavioral management options have been implemented. Ultimately, dental treatment under general anesthesia will have greater benefits than risks.

#### **Patient perspective**

The parents thanked the authorities who took tremendous care of their children and were happy and satisfied by the support provided by the hospital authorities and staff in taking care of their child. Furthermore, they were satisfied by the outcome of the treatment which improved the patient's discomfort and emotional well-being.

#### Informed consent

Informed consent was obtained for the procedure from the parents before the procedure under general anesthesia.

#### **Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parent (s) has/ have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patient's parents understand that their his names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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# **Conflicts of interest**

There are no conflicts of interest.

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