

Case Report

Dental management of tricho-dento-osseous syndrome in adolescent patients: Literature review and case presentation

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ABSTRACT

Tricho-dento-osseous syndrome (TDO) is a rare autosomal dominant disorder with complete penetrance. Common clinical features include abnormalities of hair, teeth, and skull. Dental management of TDO patients is quite challenging in terms of existing dental and skeletal problems. The current article presents a 12-year-old girl suffering TDO, followed by a review on the published literature pertaining to the dental management of TDO patients. Patient history included, rejected corneal transplantation, stone-forming kidneys, and several previous dental treatments. She was noted to have signs of mandibular prognathia, frontal bossing of the skull, mild bilateral tibial bowing, microstomia, and labial fissures. Dental findings included severe generalized enamel defects, discolored teeth, microdontia, anterior open-bite, posterior cross-bite, deep periodontal pockets, hyperplastic inflamed gingiva, taurodontism of permanent molars, dental periapical radiolucencies, and missing teeth. She was the only child of healthy, nonconsanguineous parents with no familial history of similar congenital syndrome or dental abnormalities. A treatment plan was established based on medical/dental history and findings, using a team-based approach. This article emphasizes the importance of a multidisciplinary approach for the dental management of patients suffering TDO.

Key Words: Adolescent, dental enamel hypoplasia, taurodontism, tricho-dento-osseous syndrome

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INTRODUCTION

Tricho-dento-osseous syndrome (TDO) is a rare autosomal dominant disorder with complete penetrance.^[1] Common clinical features include abnormal hair (kinky, curly, and brittle), enamel defects (hypoplasia, hypocalcification, and hypomaturation), hypodontia, taurodontism of both primary and permanent teeth, skeletal abnormalities (thickening of skull bone, sclerosis of the skull base, tubular sclerosis, and obliteration

of the frontal and mastoid sinuses), finger clubbing, and brittle fingernails.^[2-6] A study by Nguyen also revealed that patients affected by TDO showed class III skeletal pattern, longer mandibular corpus length, and shorter ramus height.^[7]

Several different mutations in homeobox 3 gene (*DLX3*) has been identified in individuals with TDO.^[8-10] Considering the variability of features, efforts have been made to categorize TDO into

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subtypes. Some researchers reported the same genetic bases despite different phenotypes.^[3] Although, a recent study has shown different genetic variants associated with different phenotypic presentations.^[11]

Dental management of TDO is challenging due to several reasons such as technical difficulties of endodontic treatment of molars with taurodontism, lack of appropriate abutment for prosthodontic and orthodontic appliances, disturbances in tooth eruption, and behavioral management of young patients needing complicated treatments at early age.^[12,13]

Although there are several articles reporting TDO, scant attention has been paid to the management of dental problems. The reported case is also remarkable because it shows some uncommon manifestations on the syndrome. Thus, the purpose of this literature review and casereport is to describe manifestations and dental treatment approaches to TDO and discuss the challenges of comprehensive team-based dental treatment.

CASE REPORT

A 12-year-old girl was seen in the Pediatric Department at Dental School of Mashhad University of Medical Sciences. She was referred by a general dental practitioner, requesting comprehensive dental management. Her last visit to dentist was about two years earlier with the chief complaint of bad anterior teeth.

She had been diagnosed with TDO syndrome, 7 years previously. She was the only child of healthy, nonconsanguineous parents. She had been delivered naturally at term with a normal birth weight. The diagnosis had been made by a team of qualified specialists (pediatric developmental specialist, pediatric endocrinologist, and radiology specialist), based on history, physical examinations, radiographic findings, and laboratory tests. Genetic investigations to find DLX3 mutation was not carried out. There was no familial history of similar congenital syndrome or dental abnormalities. Thus, the case was considered to be the product of a sporadic mutation.

The patient had normal intellectual abilities. Despite being shy, she was eager to talk about her educational goals, hoping that her achievements would compensate for her physical shortcomings.

Past medical and dental history

During the past year, the patient had received a corneal transplantation of the right eye as a treatment

for idiopathic corneal ulceration. Unfortunately, the transplanted cornea was rejected.

She had also been diagnosed to have stone-forming kidneys. Several stones were successfully dissolved, and the patient was receiving 500 mg D-Penicillamine daily as a prophylactic measure. History also revealed mild congenital heart disease (a ventricular septal defect which was successfully closed surgically during the first years of her life) for which she was not receiving any medication or treatment, as decided by the cardiologist.

According to previous dental records, she had a history of pulp necrosis and multiple dental abscesses. Endodontic treatment of teeth numbered 11, 21, 22, 36, 32, 42, 42, and 46 (FDI system) had been performed about two years ago. Tooth number 42 had been restored using a fiber post. Restorative treatment of teeth numbered 16, 15, 26, 36, and 46 had been also performed using prefabricated stainless steel crown. Teeth numbered 11, 21, 22, 32, 42, and 42 had been restored using composite resin material.^[14]

Physical and dental examinations

Although the patient could walk with no problem, a mild bilateral bowing of legs was detectable. She was noted to have signs of mandibular prognathia and frontal bossing of the skull. Her hair and nails were normal, but her skin was rather dry. Microstomia, labial fissures, and rejected corneal transplant were also notified [Figure 1].

Intraoral examinations revealed severe generalized enamel defects affecting all teeth. Other findings included discolored teeth, microdontia, skeletal and dental class III malocclusion, anterior open-bite, posterior cross-bite, deep periodontal pockets (teeth numbered 15, 11, 21, 22, 25, 26, 31, and 41), marginal gingival inflammation of maxillary anterior teeth, and hyperplastic inflamed gingiva covering teeth numbered 23, 24, 34, 33, and 44 [Figure 2]. At that time, she reported no dental pain. There were several previous restorations (as discussed in section 2–1) some of which needed to be repaired.

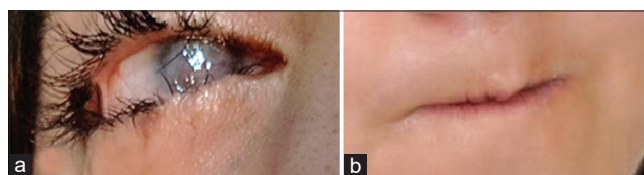


Figure 1: (a) Rejected corneal transplant (b) Microstomia and labial fissures.

Radiographic examinations

No abnormalities were found on radiographs of the hips, vertebrae, and upper limbs. The radiologist reported a mild bilateral tibial bowing. Main features revealed by dental panoramic radiography were as follows: taurodontism of permanent molars, forcation radiolucency in tooth 47, periapical radiolucencies in the anterior mandibular region and around the apex of tooth 26, and missing mandibular central incisors (teeth numbered 31 and 41) [Figure 3].

Treatment plan and interventions

Individual treatment plan was established using a team-based approach. Patient and parents were instructed to improve routine oral hygiene practices and also were instructed to apply home-based primary/preventive modalities including alcohol-free 0.05% sodium fluoride and 0.12% chlorhexidine mouth rinses, and casein phosphopeptide-amorphous calcium phosphate (CPP-ACP) paste. Thorough dental prophylaxis was performed using rotary prophylaxis brush and nonabrasive prophylaxis paste. Professional application of 5% sodium fluoride varnish was performed every 3 months. Oral hygiene compliance and dental condition were assessed in scheduled monthly visits for the first 6 months and 3-monthly intervals afterward.

Regarding the low rate of pulp treatment success in TDO,^[15] prophylactic endodontic treatments were avoided. Tooth number 26 which was considered periodontally hopeless due to extensive pulpoperiodontal lesion was extracted. Endodontic treatment of tooth 47 was performed due to idiopathic pulp necrosis and was covered with prefabricated stainless steel crown. Anterior composite restorations of lower incisors were repaired. Full debonding of the restoration of tooth 42 was observed at 6-months recall probably because of difficulty in maintaining a dry field and poor quality and quantity of remaining dental tissue, so the tooth was temporary restored using a prefabricated polycarbonate crown until a definitive custom-made crown could be used. At 12-months recall, restoration of tooth 32 was also debonded [Figure 4].

Furthermore, the patient had esthetic concerns about her smile. Despite having complex orthodontic problems such as malposed teeth and excess space, the team decided that orthodontic treatment was not possible. Bond failure of orthodontic brackets to the defective enamel and presence of teeth that



Figure 2: Intraoral frontal view.



Figure 3: Initial dental panoramic radiograph.



Figure 4: Mandibular teeth at 12-months-recall.

were partially or totally covered with gingiva were factors contraindicating orthodontic treatment. The team also concluded that the remaining teeth were not inappropriate conditions to support fixed prosthesis. A removable overdenture for the upper arch was fabricated in order to obtain acceptable aesthetic and functional results, and her smile without embarrassment was seen for the first time [Figure 5].

Furthermore considering the high risk for developing psychological problems, the patient was referred for psychiatric consultation, although during the next



Figure 5: Interim maxillary overdenture.

recalls, parents stated that they did not feel the need for consultation.

DISCUSSION AND REVIEW OF LITERATURE

TDO is a rare congenital syndrome mainly affecting hair, teeth, and bones. Involvement of other organs such as flat and/or brittle fingernails, clinodactyly, and skin lesions are also reported.^[13] In the present case, further uncommon features including stone-forming kidneys and idiopathic corneal ulceration were observed. Defective enamel is considered the hallmark of the syndrome, and other phenotypes such as hair problems may be mild or absent, as in the present case.^[3]

Several factors make dental treatment for TDO patients a challenge for dentists. Psychological considerations, challenges of endodontic treatment of taurodonts, and problems associated with bonding of adhesive restorative materials to defective enamel are some of these factors. Different aspects of treatment planning for TDO patients are discussed next.

Psychological considerations

In patients suffering TDO, dental treatments usually start at an early age. Although craniofacial bones are significantly affected, intellectual capabilities remain untouched.^[16,17] and simple preventive or treatment interventions can be performed using routine behavior management techniques. Complex dental treatments may need to be performed under general anesthesia or sedation (as in normal young or uncooperative patients).

Numerous psychological problems are associated with craniofacial anomalies in children and adolescents.^[18,19] Besides craniofacial anomalies, dental phenotype of amelogenesis imperfecta (AI)

which is similar to TDO, is associated with higher levels of distress and social avoidance.^[20] Having normal intellectual abilities, children gradually become aware of their different appearance and would be at great risk of psychological problems such as depression, low self-esteem, and social isolation. Furthermore, regarding psychological adjustment to craniofacial anomalies, children and adolescents with syndromic anomalies have lower scores than nonsyndromic anomalies, revealing a deeper problem associated with syndromic conditions.^[21]

The following strategies can be applied by the dentist in order to reduce psychological problems:

- Referring the patient to a mental health professional:
It is wise for the dentist to refer child and adolescent patients suffering syndromes with negative influence on esthetics (including TDO), to a mental health professional. Establishing appropriate psychological counseling, diagnosis, and therapies are normally beyond the dentist's capability.
- Avoiding unrequested attention as much as possible:
Patients with facial aesthetic problems, usually feel uncomfortable with behaviors such as staring, asking about, or remarking on their condition.^[22] Thus, the dentist should avoid such behaviors as much as possible, and the patient's history should be obtained from parents (prior to the patient's first visit).
- Scheduling appointments for patients with similar conditions:
When in physician's or dentist's waiting room, people tend to talk and share their experiences. Meeting other parents and children in similar situations, helps them feel that they are not alone.^[23,24] Scheduling appointments for patients with similar conditions to TDO (e.g., other congenital craniofacial anomalies, AI, and cleft lip/palate) *in close proximity* brings the opportunity for parents and children to communicate.
- Intermediate esthetic treatments:
Despite the fact that performing definitive esthetic dental treatments is usually not possible in adolescents, intermediate treatments can improve their appearance and self-confidence.

Endodontic considerations

Considering the fact that successful endodontic treatment of taurodontism has rarely been reported,^[13] thorough follow-ups are crucial and failure might be

expected, especially for immature teeth with thinner dentinal wall and open apex. The decreased success rate of endodontic treatment of taurodonts may be related to the wide variation in root canal configuration and pulp chamber anatomy, different degrees of pulp canal obliteration, increased possibility for extra root canals, and apically positioned orifices.^[25]

Another endodontic challenge for TDO patient is early occurrence of pulpal disease while the apex is still open. Having a weak enamel, the teeth in TDO patients are more susceptible to dental caries and attrition which can lead to pulpal exposure, periapical lesions, and the need for endodontic treatment at young age. Pulp treatment of open apex teeth may result in the need for apexification, apexogenesis, or regenerative endodontic treatments. Considering the challenges mentioned above surrounding endodontic treatment of taurodonts, vital pulp therapy instead of full pulp extirpation is the treatment of choice.

In addition, idiopathic pulp necrosis occurred in tooth number 47 in the present case despite being partially covered with gingiva. A possible theory leading to pulp necrosis of the tooth is preeruptive coronal resorption which was confirmed by clinical exploration. Pre-eruptive coronal resorption has been reported in several cases of AI.^[26-28]

The following advices may decrease the risk of endodontic failure: Careful exploration for additional orifices and canals using magnification:^[24,25]

- Adequate irrigation and root canal disinfection:
Repeated irrigation with 2.5% sodium hypochlorite, and the use of ultrasonic irrigation.
- Using a combination of root canal filling techniques:
lateral compaction technique in the apical region and vertical compaction technique in the pulp chamber.
- Vital pulp therapy in young permanent teeth:
For teeth with open apices, vital pulp therapy instead of full pulp extirpation is the treatment of choice.
- Providing a perfect coronal seal after root canal treatment:
A full-coverage technique is preferred.
- Failure of endodontic treatment is highly expected, thus close follow-ups are crucial for early detection.
- Preservation of the alveolar bone for definitive future prosthetic or implant treatments:

Attempt should be made to maintain teeth with poor long-term prognosis via endodontic treatment.

Restorative considerations

Because of rarity of TDO and the fact that dental problems are the most consistent features of the syndrome, the condition is commonly mistaken for AI. Although limited evidence is available regarding restorative treatment is TDO, a satisfying amount of literature is available relating to AI.

As with AI which is a condition of somehow the same origin with TDO,^[29] different restorative materials are suggested such as conventional or resin-modified glass ionomer cements, compomers (polyacid-modified resin composites), resin composites, and indirect adhesive restorations (cast onlays or crowns); but none of them have been proven to be the best.^[29,30]

The use of direct restorative materials such as amalgam or adhesive resins in TDO is quite challenging. As with AI, TDO teeth are at greater risk of marginal fracture because of the defective dental tissue surrounding restorations. Amalgam restorations can be used successfully in mildly affected posterior teeth.^[31,32]

Direct resin composite restorations for esthetic zone are recommended to restore esthetics and prevent attrition of anterior teeth in young patients. Restoration using adhesive materials (composite resin and glass ionomer) have occasionally been reported to be more successful than amalgam restorations.^[31,33] However, studies have declared a reduced longevity of adhesive restorations in AI compared to control group. Placement of more definitive restorations (such as porcelain veneer or full crown) should be postponed until the tooth is fully erupted. Young patients should always be informed of the likely need for repair or replacement of the restorations.

The reduced longevity of adhesive restorations could be best explained by poor quality and quantity of enamel which leads to inferior etching pattern and insufficient bonding area as less enamel and more dentin is engaged. Decrease in bond strength results in increased marginal leakage, lower retention, and reduced longevity of adhesive restorations.^[33,34]

Several modifications to conventional adhesive systems have been suggested in order to improve the bonding quality in AI (such as deproteinization of enamel using sodium hypochlorite), but the results were not promising.^[35-37]

Full coronal coverage of primary (final restoration) and permanent (interim restoration) molars using prefabricated stainless steel crowns is recommended in young patients with TDO, in order to prevent dental caries, pulp infection, attrition, and decrease in vertical occlusal dimension.^[13,38,39] Care should be taken to perform minimal if any, preparation preceding crown placement.

It is suggested to avoid the use of endodontic posts for prosthesis retention in permanent molars affected with taurodontism. It is also recommended not to use taurodonts as abutments for either prosthetic or orthodontic appliances.^[40] However, partial or complete overdenture can be considered as an interim potential treatment for young patients. Overdentures can preserve alveolar bone around remaining roots and teeth which is essential for future definitive treatments such as dental implants.

As a summary, the following recommendations are advised for restoration of TDO teeth in adolescents:

- Teeth of the esthetic zone:
Interim composite resin restoration is performed until definitive treatment can be performed.
- Posterior teeth:
Mildly affected teeth can be restored by direct restorative materials (either composite resin or amalgam) and provisional stainless steel crown restoration is placed for moderately or severely affected teeth until definitive treatment can be performed.
- Use of overdentures:
Partial or complete overdentures can be used as interim treatment until a definitive treatment can be performed.
- Close follow-ups:
Considering lower success rate of restorative treatments in TDO, close follow-ups are recommended to assess the need for repair or replacement.

Periodontal considerations

There is limited literature discussing periodontal status of TDO patients. Although teeth with taurodontism seem to be at lower risk of periodontal diseases due to the more apically placed apex and are usually in normal periodontal condition.^[25,40] this may not apply to TDO patients who are affected by coexisting problems.

Although periodontal status in patients with disorders such as AI does not seem to differ from normal

patients,^[31] they are more susceptible to dental caries and periodontal diseases due to the need for prosthetic and restorative treatments at an earlier age.^[41] Having multiple partially impacted teeth which are reported as a feature of TDO^[42,43] may also facilitate gingival infection, formation of periodontal pockets, and pericoronitis.^[44,45] Therefore, thorough examinations of periodontal condition should be performed in conjunction with prophylactic or, if needed, periodontal treatments. Furthermore, periodontal health affects the success of the restorative treatments while periodontal regression might lead to exposure of restoration margins and need for further treatments.^[46]

Preventive considerations

TDO patients are at a greater risk for dental caries and tooth loss due to several factors, thus, stricter preventive measures are needed to maintain oral and dental health. Delivering instructions on oral care, plaque control, and personal dietary advice should be reinforced and monitored on repeated occasions.

Prescription of self-applied dental care products such as alcohol-free sodium fluoride and chlorhexidine mouth rinses, CPP-ACP paste is recommended in order to prevent dental caries and even reverse or stop lesion progression. The use of desensitizing fluoride toothpastes (by a soft toothbrush) and products containing potassium nitrate is also indicated in case of tooth sensitivity.^[13,47-49]

Patients should receive both standard and tailored dietary advice including reduced intake of sugary or acidic food and beverages (e.g., sour foods, citrus juice, cola, and any carbonated drink), encouraged consumption of alkaline or neutral foods (e.g., legume, milk, and water) and use of xylitol-containing chewing gums.^[13,50-52]

CONCLUSION

Dental management of patients with TDO is a long time process starting at an early age. Although tooth loss might be inevitable, saving the teeth for as long as possible is important for an appropriate craniofacial and psychological development as well as maintaining alveolar dimensions. Achievement of this goal requires collaboration of a multi-disciplinary team of professionals.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the

patient and her caregiver has given consent for her images and other clinical information to be reported in the journal. The patient and her caregiver understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

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