Esophageal Atresia with Tracheoesophageal Fistula and Gastroesophageal Reflux in Children: Dental Considerations and Case Report

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Background: Esophageal Atresia (EA) is defined as the congenital interruption of the continuity of the esophagus. Pediatric patients also have other congenital conditions, such as Tracheo-Esophageal Fistula (TEF). **Case report:** A 7-year-old male with TEF referred by a Pediatric Cardiologist, with the principal complaint of "severe and generalized tooth wearing". Considering that the patient was systemically stable, it was decided to perform the oral procedures under local anesthesia and rubber-dam isolation with an antimicrobial prophylaxis regimen. The treatment consisted of the extraction of all maxillary primary incisors and canines and both first molars; in the mandibular arch, only the lower second right molar was extracted, and a distal shoe was placed. Pulpotomies were performed and preformed metallic crowns were placed on the remaining second primary molars, on both lower first molars, and on lower canines and lateral incisors. Finally, a fixed prosthesis was positioned in the upper arch, and cemented through orthodontic bands adapted to both crowned second molars. The patient has been maintained under close medical and dental control. The child showed satisfactory oral conditions, and the vomiting episodes had decreased significantly. **Conclusions**: Dentists can learn and then participate in the integral health management of infants and young children affected with EA/TEF, particularly those with dental erosion.

Key words: Esophageal Atresia, tracheo-esophageal fistula, dental management, children, clinical case.

INTRODUCTION

Begin a sophageal Atresia (EA) encompasses a group of anomalies in newborns and infants, comprising the narrowing or peristalsis of esophagus, and, in severe cases the congenital absence of the continuity of the esophagus' trajectory toward the stomach.¹⁻³ EA combined with Tracheo-Esophageal Fistula (TEF) causes in newborns an inability to feed and swallow, together with respiratory problems and gastrointestinal reflux. When the newborn swallows, milk is unable to pass through the esophagus tract to

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reach the stomach, and thus essential nutrients are not well digested and absorbed; another potential adverse event occurring when bottle feeding and swallowing is the liquid diffusion into the trachea and inward the lungs, which may cause pneumonia.¹

EA/TEF is the most common congenital anomaly of the esophagus in young children, presenting a multifactorial etiology.⁴ This non-familial disease occurs in around one in 2,500–4,000 live births, with a slight predominance of males; once a mother has a child with EA, the risk of having a second child with the anomaly increases to 1%. Over 50% of patients also have other congenital conditions, such as *Vertebral*, *Anorectal*, *Cardiovascular*, *Tracheo-Esophageal*, *Renal*, and *Limb* defects (together constituting the acronym *VACTERL*, which has been taken as a syndrome of associated anomalies).^{3,5}

The majority of cases of EA is sporadic/non-syndromic, although there is a small number of cases associated with chromosomal abnormalities, including Trisomy 13 (Edwards' syndrome), Trisomy 18 (Bartholin-Patau syndrome), and Trisomy 21 (Down syndrome).^{2,5,6} Diagnosis of EA/TEF is commonly made during the first hours of life, by means of observing the typical symptoms, or when a small or absent stomach bubble is exhibited in a plain x-ray of the chest and abdomen; a primary sign, when EA is suspected, is the impossibility of passing an orogastric catheter (or

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a tube) through the esophagus beyond 11 or 12 cm.⁷ Reparative and definitive surgical treatment consists of disconnection of the TEF, closure of the tracheal defect, and anastomosis of the esophagus to the stomach.⁸

Improvement of the survival and Quality of Life (QOL) of children suffering EA has increased over the last four decades. This is largely attributable to advances in neonatal intensive care, anesthesiology techniques, ventilatory and nutritional support, antibiotic regimes, opportune surgical interventions, and improved surgical materials and techniques.^{1,3} Due to this increase in survival, the morbidity associated with EA/TEF treatment has become an important public-health issue.⁷

Oral health implications –for example, dental erosion associated to gastroesophageal reflux disease GERD– and management considerations in pediatric patients have been scarcely covered in the literature. The aim of the present report was to describe a case of a 7-year-old boy and the dental treatment provided to him.

Case report

A 7-year-old male presented with his parents at the Pediatric Dentistry Clinic, requesting dental review and treatment. The child was referred by a local Pediatric Cardiologist, with the principal complaint of "severe and generalized tooth wearing".

Medical history. The parents (father, aged 28-years, and mother, aged 32-years) reported that polyhydramnios was detected during pregnancy, and negative history of parental drug taking, or exposure to infections, trauma, or irradiation. Labor occurred at the fulllength pregnancy and was uneventful. The patient initially displayed apparent good general health and an asymptomatic status. At birth, the infant's weight (2,290g) and height (42cm) were considered to be below the population median, according to the Mexican chart of newborn's growth. The baby was kept at home until the 3rd day of life and was breast-fed, despite repeated gastroesophageal reflux, vomiting, regurgitations and respiratory problems. Then, on day 5 after birth, the patient was taken to a primary-care pediatrician, who made the diagnosis of EA-TEF with pneumonitis; the child was submitted to a surgical intervention, performed on day 7 of life. Post-operatively, nasogastric feed was started from day 2 onward, and breastfeeding was allowed from day 7. The patient was re-intervened at 12 and 18 months of age, intending the surgical TEF closure, without achieving proper repair. Thus, a gastrostomy catheter was inserted to feed the patient (Fig. 1). However, and for around 6 months, the mother insisted on feeding her son by mouth; vomiting episodes persisted, and the child started to lose weight.

Craniofacial examination. The child displayed some of the typical features of "long face syndrome": an asymmetric hyperdivergent face, convex profile, increased lower anterior-face height, eye bags, slight chin deficiency, gummy smile, and evident oral breath with lip incompetence (Fig. 2).

Intraoral examination. The oral cavity exhibited mixed dentition with evident generalized loss of dental structure by erosion, in all upper arch teeth, and in the lower primary molars and canines. Permanent lower central incisors were well erupted, but the cusp tips of the four first permanent molars were just emerging into the oral cavity. In addition, the patient exhibited very poor oral hygiene with noticeable plaque accumulation. Deep occlusal caries lesions were also observed in both lower second primary molars. Upper incisors, canines, and first molars exhibited severe dental erosion with total coronal destruction. The majority of teeth had concomitant enamel hypoplasia/hypomineralization pigmentations and white spots (Fig. 3). Halitosis was notorious. Occlusal evaluation detected an obvious anterior open bite (of around 10mm), caused by a combination of thumb sucking, tongue thrusting, and oral breathing; there were no interdental spaces, and a bilateral straight terminal-step relationship between primary second molars was observed.

Figure 1. Face frontal view.



Figure 2. Abdominal gastrostomy catheter.



Figure 3. Initial intraoral views.



Treatment

Based on the information collected, the patient's physical status, the consultation with the Pediatric Cardiologist, and considering that the patient was systemically stable, it was decided to perform the oral procedures under local anesthesia and rubber-dam isolation. An antimicrobial prophylaxis regimen was provided; it consisted of Amoxicillin 50 mg/kg orally, 30 min before the start of dental procedures. The parents fully understood and agreed to the planned dental treatment by means of a signed, written informed consent. The treatment consisted of the extraction of all maxillary primary incisors and canines and both first molars; in the mandibular arch, only the lower second right molar was extracted, and a distal shoe was placed. Pulpotomies were performed and preformed metallic crowns were placed on the remaining second primary molars, on both lower first molars, and on lower canines and lateral incisors. Finally, a fixed prosthesis was positioned in the upper arch, and cemented through orthodontic bands adapted to both crowned second molars (Fig. 4). Total duration of dental treatment was 7 weeks. Also, instructions for an exhaustive and closely monitored hygiene/nutritional program were provided; the program consisted of frequent professional dental cleanings, topical fluoride therapy, and tooth brushing and chlorhexidine mouth rinses at home. Additionally, the abnormal oral breath was managed by a Pediatric Otorhinolaryngologist. Since then, the patient has been maintained under close medical and dental control. In the last dental examination, the child showed satisfactory oral conditions, a good oral hygiene level and, according to his parents, the vomiting episodes had decreased significantly. At present, a new surgical TEF closure is pending; it will be performed when the patient returns to his normal weight.

DISCUSSION

The proper management of pediatric patients affected with EA/ TEF remains as one of the challenging issues in dentistry. This disorder requires the early involvement of a multidisciplinary pediatric health care team, consisting mainly of a Neonatologist, Cardiologist, Surgeon, Nutritionist, Otorhinolaryngologist, Occupational Therapist, and Psychologist. In many cases, the Pediatric Dentist can play an important role in maintaining the oral cavity in a healthy and functional condition, free of local infectious or inflammatory processes.¹ The orofacial consequences of EA/TEF in newborns and infants should be carefully considered by practitioners, particularly

Figure 4. Post-dental treatment intraoral views.



by those who provide oral health services in hospitals.^{9,10} For example, special care is provided if newborns cough, choke, vomit, or the skin turns blue while feeding. Also, feeding and breathing difficulties in early infancy may influence the growth and development of the orofacial region, and, when sensory motor skills are present, the child may exhibit troubles in performing adequate tooth brushing and flossing.¹¹

EA with or without TEF is a neonatal emergency that requires surgical repair for the patient survival. This depends on stabilization and a proper resuscitation followed by appropriate post-operative care. Esophageal surgery can be conducted in all age groups. Surgical treatment is considered urgent, but not an emergency.⁷ Thus, it is preferable to perform the intervention during the first days of life, provided that there are not pulmonary or other major anomalies.8 Even after surgical closure of the TOF, patients may develop scar tissue that can make swallowing problematic, or partially block the food passage to the stomach.3 Likewise, around 50% of children who underwent surgical repair continue to suffer gastrointestinal reflux, causing the return of gastric acids from the stomach, and consequently a burning pain denominated "heartburn"; in addition, the risk of producing severe dental erosion is high,12-15 as occurred in our patient. However, other authors have not found evidence of the relationship between GERD and dental erosion in these patients.¹⁶

Because of the controversy that loss of primary tooth structure by erosion is associated with gastroesophageal reflux in children with EA/TOF, we decided to perform an exhaustive dental literature search to retrieve those relevant articles on this specific clinical topic, the most important in for the pediatric dentist, published since the year of 1990. Two electronic databases were explored (PubMed and EMBASE), and as a result, 15 studies could be selected and analyzed by the authors. Methodological characteristics and main reported clinical findings from these articles are described in the Table 1. Other practical and clinical measures to consider and implement in children with EA/TEF are included in Table 2.

Authors, year (country)	Design, aims, and methodology	Main clinical findings
Taylor et al. 1992 (US) ¹⁷	A clinical case report of an 8 year old boy with symptoms of GERD. Extensive loss of structure in primary teeth and the four first permanent molars.	The child was treated symptomatically with metoclo- pramide to reduce the volume and frequency of acid gastric content, and ranitidine to suppress excess excessive gastric acidity. He received twice daily applications of neutral sodium fluoride gel at home. Sodium bicarbonate rinses were also recommended.
Aine et al. 1993 (Finland) ¹⁸	A cross-sectional study. 15 GERD children (median age 8.1 years). Dental examinations for dental erosion. Plus 2 clinical case reports.	Erosive lesions were found in 13 of the 15 children diagnosed with GERD. Young and school-age children with generalized dental erosion, even though in apparent good health, should be referred to the Pediatric gastroenterologist.
Dodds et al. 1997 (US) ¹⁹	A case report of a 3.25 year-old girl suffering GERD. Counselling on oral hygiene and diet. Dental treatment included prophylaxis, topical application of 1.23% acidulated phosphate fluoride, and restorations with glass-ionomer cement.	Conservative management of GERD includes the averting of carbonated beverages, spicy foods, tea, coffee, and decreasing dietary fat. Furthermore, sleeping in a prone position should be avoided, as is eating within 3 hours of retiring. Antiacid drugs to increase pH and deactivate pepsin are the first line of pharmacological agents. Dental management should include dietary counselling, daily topical fluoride applications, sodium bicarbonate mouth-rinses, pit-fissure sealants, adhesive resto- rations, and preformed crowns.
O'Sullivan et al. 1998 (UK) ²⁰	A cross-sectional study to investigate the relationship between dental erosion and GERD in 53 children aged 2-16 years.	Acid reflux has been shown to reach different levels of the esophagus, with a 4 to 5-fold lesser acid exposure in the proximal esophagus compared with the distal esophagus. 17% of participants showed any sign of dental erosion, and only one of them had erosion involving dentine. These results suggest that dental erosion may not be a great problem in children, as it is in adults.
Linnett et al. 2001 (Australia) ²¹	A narrative literature review. Prevalence, clinical manifestations, etiology of dental erosion in children. Guidelines on preventive and restorative options.	The prevalence of dental erosion in children varies between 2 to 57%. Symptoms of dental erosion range from sensitivity to severe pain associated with pulp exposure. Regurgitation of gastric contents into the oral cavity, as occurs in GERD, is the most common cause of dental erosion. Dental rehabilitation involves stainless steel crowns to restore lost vertical dimension, and composite resins for aesthetics.
Linnett et al. 2002 (Australia) ²²	A comparative clinical and in vitro cross-sectional study on oral health status and dental erosion. 52 pairs of GERD and matched controls. Dental examinations and salivary samples for <i>S. mutans</i> .	The prevalence of dental erosion was statistically higher in GERD children (14%) in comparison to healthy controls (10%). Caries experience was also higher in affected children. No statistical difference between groups regarding salivary <i>S. mutans</i> counts (42% in GERD participants vs. 25% in healthy participants).
Ersin et al. 2006 (Turkey) ²³	A cross-sectional study. Aim: To investigate the effects of GERD on erosion, caries formation, salivary function, and salivary microbial counts. 38 GERD and 42 healthy children (mean age of 6.5 years old).	Prevalence of dental erosion and <i>S. mutans</i> counts were found to be significantly higher in GERD children. Caries experience, salivary flow rate and buffering capacity, and frequency of acidic drinks, foods, and sugar consumption were found to be similar in both groups.
Pace et al. 2008 (Italy and US) ²⁴	A systematic review. Justification: The real impact of GERD in the genesis of dental erosion remains unclear. The search was carried out in PubMed and the Cochrane Controlled Trials Register. 17 studies (5 in children) met the selection criteria.	There is a strong association between GERD and dental erosion. Children with GERD are found by the majority of studies at increased risk of developing dental erosion in comparison with healthy subjects, as are being disabled people. Child's oral cavity inspection in search for dental erosion should become a routine practice in patients with GERD.

Table 1. Methodological characteristics and main reported clinical findings extracted from the selected articles.

Authors, year (country)	Design, aims, and methodology	Main clinical findings
Dahshan et al. 2012 (US) ²⁵	Cohort study. To evaluate the presence of GERD and dental erosion in 24 children with primary/permanent dentitions. 2-18 years old.	20 patients exhibited dental erosion. The pattern of dental erosion involved mostly primary posterior teeth. Erosions involving primary teeth may not progress to involve the permanent teeth if identified early and the causal factors adequately treated.
Taji et al. 2010 (Australia) ²⁶	A narrative and critical literature review on dental erosion in children, on regard to its etiology, prevalence, associ- ated clinical conditions (GERD), and prevention.	The prevalence of dental erosion among GERD children ranges from 10 to over 80%. Gastric hydrochloric acid has a pH of 1-1.5. While diurnal reflux could be damaging, reflux during sleep cause the greatest loss of dental hard tissues, presumably due to the reduced saliva flow.
Wild et al. 2011 (US) ¹⁶	Cross-sectional study. 59 children with symptoms of GERD and 20 healthy chil- dren controlled by age, dietary intake and oral hygiene. 9-17 years old. Prevalence of dental erosion among participants.	The proportion of teeth affected by dental erosion was similar in symptoms compared with asymptomatic children. Location-specific dental erosion is not associated with GERD, salivary flow, or bacterial load.
Firouzei et al. 2011 (Iran) ²⁷	A systematic review. Association between dental erosion and GERD. All descriptive or analytical studies up to December 2011. PubMed and Scopus electronic databases.	15 studies were selected, 5 in child populations. There was a strong association between dental erosion and GERD, but relationship in children was found to be of less importance. Early diagnosis, treatment, lifestyle changes, and medications can prevent further damage and tooth loss.
Monagas et al. 2012 (US) ²⁸	Cross-sectional study to assess the prevalence of upper gastroesophageal symptoms and to test for GERD in children with dental erosion. All patients (3-21 years old) referred by dentists for GERD evaluation because of dental erosion.	There were 16 participants (15 male) with GERD, aged 5-12 years. Of them, 11 had upper gastroesophageal symptoms or abnormal tests. The authors found upper gastrointestinal signs or symptoms in the majority of participants with dental erosion. However, there was no correlation between the stage of dental erosion or the number of affected teeth and GERD.
de Oliveira et al. 2016 (Brazil) ²⁹	Case-control study. To investigate the impact of GERD on dental erosion, and the association between dental erosion with diet, oral hygiene, and socio-demographic characteristics. 43 GERD children and 136 paired controls, 2-14 years old.	Dental erosion was detected in 10.6% (25. 6% in GERD children and 5.9% in controls), and it was not associated with dietary consumption or socio-demo- graphic characteristics. GERD children who used adult toothpaste had a 5.8 higher chance of having dental erosion.
Ganesh et al. 2016 (US) ³⁰	A prospective cohort study in 27 children aged 3 years or older. To determine the relationship between dental erosion and acid and non-acid GERD, measured using a combined pH and multichannel intraluminal impedance.	The prevalence of dental erosion was 37%. There was a positive correlation between acid reflux parameters and dental erosion. Acid, rather than no-acid reflux, seems to have a significant role in dental erosion.

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CONCLUSIONS

In conjunction with the Gastroenterologist, the Psychologist and the Occupational Therapist, Pediatric Dentists can learn and then participate in the integral health management of infants and young children affected with EA/TEF, particularly those with dental erosion, by maintaining the chewing and swallowing skills, minimizing sensory abnormalities, and providing playful and pleasurable nurturing experiences. Practitioners may collaborate early in preparing parents for feeding-related difficulties and for providing guidance for improving oral sensory motor skills, thus, the social rapport of the patients' upcoming future life.

Table 2. Medical and dental considerations in children with EA.

Medical considerations	Dental considerations
The affected baby should be nursed with the chest in an upright position.	Discuss the dental treatment plan with the child's primary physician.
Children with cardiac malformations should be carefully evaluated and the Pediatric Cardiologist should be consulted to consider an antibiotic prophylaxis regimen before dental treatment.	Dental erosion can be treated using rehabilitation procedures with minimal intervention, for example, adhesive restorations with biomimetic materials.
As part of clinical anamnesis, practitioners should focus on the presence of co-occurring disorders associated with EA/TEF.	Natural tooth structure should be preserved whenever possible. However, in cases of excessive destruction, full-coverage crowns for primary and permanent anterior/ posterior teeth should be considered.
If there are clinical manifestations of severe respiratory distress, the infant must be intubated and ventilated mechanically in an emergency room.	Chewing antacid tablets or rinses with a solution of sodium bicarbonate neutralizes the erosive effect of gastric acids on dental enamel.
In older children, non-treated patients with EA/TEF, or those with persistent problems can be trained by a therapist to learn how to suckle and breathe at the same time.	Promote an exhaustive oral preventive program, including the application of enamel remineralization agents (e.g. fluoride varnishes) and strict control of oral microflora.
There are specific medical therapies for gastrointestinal reflux: milk of magnesium hydroxide, aluminates (Maalox) histamine-2 blockers (Cimeti- dine, Ranitidine, Nizatadine), and proton pump inhibitors (Omeprazole, Lansoprazole). Consult the Gastroenterologist before administration.	
Minimize or avoid exposure to deep sedation or general anesthesia in affected children for oral rehabilitation purposes, except in strictly selected	

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