



RESEARCH ARTICLE  
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## Restorative Treatment of Amelogenesis Imperfecta Associated with Asthma: Case Report

Salim A. Algarni<sup>1\*</sup>, Sulaiman S. Alqahtani<sup>2</sup>

<sup>1</sup>Department of Restorative Dental Sciences, Faculty of Dentistry, Najran University, Najran, Saudi Arabia

<sup>2</sup>Department of Maxillofacial Surgery and Diagnostic Sciences, Faculty of Dentistry, Najran University, Najran, Saudi Arabia.

\***Corresponding author:** Salim A. Algarni, Lecturer at the Restorative Dental Sciences, Faculty of Dentistry, Najran University, Najran, Saudi Arabia. Email: saalgarni@nu.edu.sa

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### ABSTRACT

**Background:** Amelogenesis imperfecta (AI) comprises a group of hereditary disorders affecting the structure of enamel. Although many etiological factors have been implicated for the cause of these disorders, some of the systemic conditions such as asthma and dietary factors like intake of soft drinks have been correlated with AI.

**Case presentation:** This work is a clinical report of a 14-year-old girl who was diagnosed with hypoplastic type of AI. The patient had a history of asthma and she complained of dry mouth. She required replacement of her stainless steel crowns and wanted esthetic smile. In addition, she was advised composite restorations and fabrication of removable overlay upper and lower dentures.

**Conclusion:** The current case report implicated that AI could be associated with conditions such as asthma; However, future research on molecular, functional, and genetic aspects of AI integrating the inflammatory response of Asthma may help in preventing the enamel defects. Implementation of dental care for this structured population may help in preventing dental complication due to increased risk.

**Keywords:** *Associated, Treatment, Case, Report*

## INTRODUCTION

The structure of a tooth includes an enamel, dentin, and pulp, in which the enamel is a highly mineralized tissue in the human body.<sup>1</sup> However, divergence in the process of normal organogenesis can lead to hereditary disorders affecting the structure and composition of the enamel and one such disorder is amelogenesis imperfecta (AI).<sup>2</sup> The genetic alteration and frequency of occurrence have been depicted to be 1:4000 and 1:700, which are categorized according to the diagnostic criteria.<sup>3</sup> According to the Witkop classification, there are four types of AI, namely,<sup>4</sup> (i) Type I is hypoplastic enamel, (ii) Type II- hypomatured enamel, (iii) Type III – hypocalcified enamel, and (iv) Type IV- hypomatured-hypoplastic with taurodontism. The clinical characteristics of the AI typically depend on the type of defect and the stage of enamel formation. Most of the patients report destructive tooth structures, dental hypersensitivity, plaque accumulation, gingivitis, halitosis, loss of vertical dimension, and dissatisfied and compromised esthetics.<sup>5,6</sup> In addition, the mineralization of teeth, which is completed during the first 4 years of life, may have possible chances of abnormalities and association with some diseases such as asthma/bronchitis etc<sup>7</sup>: There has been an increased risk of severe demarcated opacities and destruction of tooth structures due to the use of asthmatic drugs. Several authors have reported altered salivary secretion rates, pH, buffering capacity of saliva due to poor oral hygiene and accumulating calculus and resulting pain. In addition, AI is also found to be related to factors such as socioeconomic status, age, and severity of the disorder as well the intraoral condition.<sup>8</sup> The genomic origin of AI also has been associated with syndromic conditions such as Jalili syndrome and renal enamel syndrome that could be expressed as phenotype and without oral symptoms.<sup>9</sup> Despite several etiological factors for AI, the treatment remains arduous to clinicians. It can be treated by interdisciplinary means with a combination of Oral Medicine, Periodontal,

Orthodontic and Prosthetic approaches, targeting Oral rehabilitation to the patient.<sup>8,10</sup> The clinical appearance of the patient with complaints of loss of esthetics seems to demotivate them, therefore, precise planning and appropriate treatment strategy and handling of such rare cases pose a big challenge to the dentists as they cannot guarantee relapse-free treatment. However, constant follow-up at every stage, where alteration is appreciated remains essential.<sup>11</sup> Based on the above description, we hereby report a case of AI, enlightening the clinical features, diagnosis and associated factors along with a rehabilitative treatment plan in order to strengthen the stomatognathic system of the patient.

## CASE REPORT

A 14-year-old female reported to the Oral Medicine Clinic, College of Dentistry Najran university, for the replacement of her destructive teeth, stainless steel crowns, so that she could restore her esthetics and desire for a beautiful smile. The patient reported having a history of asthma since childhood and was using inhalers containing albuterol. There was no significant family history and none of them had a similar condition. She notified a history of dry mouth for 2 years and also had appetite changes as she could not masticate owing to her destructive teeth. We conducted an unstimulated salivary flow test using the spit method and the flow was estimated to be low. She also had a habit of drinking soft drinks very often.

During the extraoral examination, there was no abnormality or dysfunction found. On intraoral examination, all the present teeth were yellowish to dark brown in color with pitting and chipped surfaces. The rough surfaces of the teeth could be observed with thin enamel. Root stumps were observed in 12, 13, 22m and the filled teeth in 33. The stainless-steel crowns were present in 14, 15, 34, 35, 44, 45, and 46. In addition, the patient presented with malocclusion, generalized attrition and periodontal examination revealed mild general gingivitis (Figures 1, 2, and 3).



**FIG 1.** Preoperative photograph (front side).



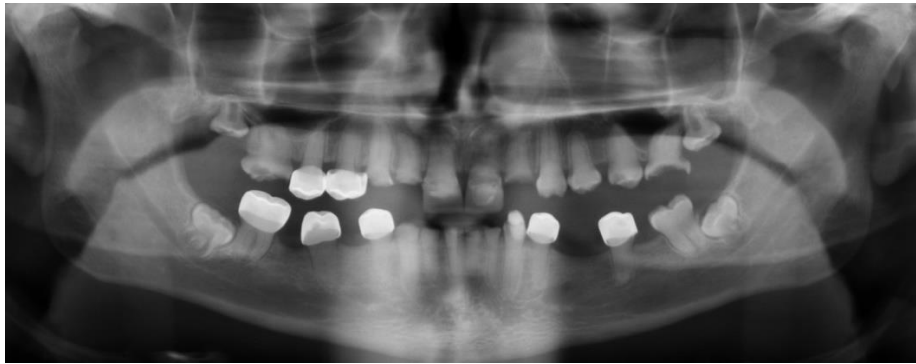
**FIG 2.** Preoperative photograph (left side).



**FIG 3:** Preoperative photograph (right side).

Radiographic examination was carried out using a panoramic radiograph and full mouth X-rays, where a thin layer of enamel was observed, but the radiodensity was greater than that of dentin. (Figure 4) Erupting tooth with open apex in 17, 27, 37, and 47 could be observed. The intraoral periapical radiographs were obtained for all the quadrants, where a thin layer of radiopaque enamel with slight obliteration of pulp chamber and loss of

cuspal height was observed along with placed crowns in posterior teeth in the maxillary and mandibular arch. Taking all the findings into consideration clinically, radiographical findings and alteration of enamel, and discoloration of teeth, the provisional diagnosis of the hypoplastic type of AI were provided. The differential diagnosis of enamel hypoplasia, secondary to dental fluorosis and dentinogenesis imperfecta were listed in our case. Although the focus was evidently pointed to the diagnosis of the hypoplastic type of AI, the treatment plan was made according to the age and patient's needs.



**FIG 4.** Panoramic radiograph.

The main aim was restoring the function and esthetics. In addition, impressions were taken for

The fabrication of removable upper and lower overlay dentures (Figure 5).



**FIG 5.** The delivery of upper and lower overlay dentures.

## DISCUSSION

AI is a group of diversity with combined hereditary and genomic origin. It has also been associated with biochemical changes and altered morphology as well as the opacities in the structure of the enamel along with altered salivation and other systemic conditions such as asthma during the mineralization and formation of tooth.<sup>6,12</sup> The four main types of AI, namely, hypoplastic, hypocalcified, hypomature, and hypomature-hypoplastic are further divided into 15 subtypes depending on the heredity and phenotype.

The collection of hereditary disorders and variations in the genes have been observed in amelogenin, enamelin, kallikrein-4, and other encoded structural proteins of enamel-such as amelobalstin, c4or f26, and enzymatic proteins such as MMP20, transcription factors such as MSX2, DLX3, cellular proteins such as WDR72, FAM83H, COL17A1, and cell receptors such as ITGB6.<sup>13</sup> Ninety percent of the proteins in the enamel matrix are amelogenin, which is expressed as AMLEX and AMLEY.<sup>14</sup>

In the present case, we observed the hypoplastic type of AI, which is caused due to failure in the secretory phase of amelogenesis, where the enamel is thin, however, we can demarcate the enamel and dentin with low contrast.<sup>6</sup> AI which is observed due to defects in the deposition of enamel matrix within the enamel is clinically seen as yellowish brown, rough, flat, smooth surfaces of teeth with/without pits, grooves, and chipping. Radiographically, the enamel lamellae appears thin, with normal density. The defects in the enamel matrix formation while differentiation of ameloblasts are appreciated in the histological sections. The subtypes of hypoplastic AI have been divided into generalized and localized pitted, diffuse smooth and rough as well as enamel agenesis.<sup>15</sup> The effects of AI have been observed in both deciduous and permanent dentition disrupting the quantitative and qualitative content of enamel.<sup>16</sup> There was no family history in our case study, hence hereditary inheritance was discarded. Although the patient provided a history of asthma since childhood, she had a habit of consuming soft drinks often. Hence, we tried to associate the terms asthma, intake of soft drinks, and dry mouth as well as the AI. The association between asthma and enamel defects has been investigated by various researchers, where in a case-control study conducted by Guergolette et al.<sup>17</sup> involving Brazilian children comparing the asthmatic patients and controls, it was shown that there was a 70% increase in enamel defects in asthmatics. Similarly, Wogelius et al.<sup>18</sup> conducted a study demonstrating the use of asthmatic drugs and the prevalence of demarcated opacities in Danish children of 6–8 years of age and found an increased risk of opacities in enamel resulting in macroscopic loss of tooth substance upon the use of beta-agonists. The explained mechanism behind this is the process of enamel formation can be sensitive to environmental disturbances, although it is genetically controlled.

Moreover, owing to sustained fever, there may be a shortage of oxygen and calcium phosphate, which in turn may disrupt the process of amelogenesis. Therefore, due to previous episodes of deprived oxygen occurring in asthmatic children, there may be an occurrence of AI, causing defects of enamel.<sup>19,20</sup> Tourino et al.<sup>21</sup> have concluded that hypomineralization and enamel defects could be observed predominantly in children who had asthma in the first 4 years of life. Similarly, Mastora et al.<sup>22</sup> conducted a retrospective case-control study to investigate the developmental defects of enamel in preschool-age children who were on asthmatic drugs and concluded severe hypoplastic lesions with loss of enamel, affecting the molars. Henceforth, the remedy might be that the pediatricians must be aware that children with asthma may have a greater chance of getting enamel defects, hence, the forthcoming complications associated with AI can be resolved. The other point of discussion is the association between soft drinks and enamel defects, where erosion of enamel has been observed due to the pH drop in the oral cavity. The acidic nature of the beverages leads to structural weakness and reduced enamel microhardness, leading to developmental defects.<sup>23</sup> The studies have reported that soft drinks with high calcium content cause less erosive changes.<sup>24</sup> The predictable factors for enamel defects can be based on the type of acid content, the titratable acidity ion concentration, and the pH value. Children have a high propensity toward soft drinks and due to the presence of caffeine, sugar, cold and acid causes high gustatory sensitivity due to the intake of beverages.<sup>25,26</sup> These consequences indirectly cause salivary dysfunction, affecting the parasympathetic innervation and damaging the salivary glands and reducing the salivary output.<sup>27</sup>

Children with oral dryness must be overlooked at the first instance as it is a clear indication for decalcification followed by low pH and reduced salivary flow.<sup>28</sup> Chair-side diagnostic tests along with obtaining detailed history and lifestyle factors are very crucial. Moreover, long-term uses of asthmatic medications have been associated with low pH and high prevalence of *Streptococcus mutans* in asthmatics. It has been attributed that mutations in several genes pave pathways and contribute to erosive tooth wear, enamel defects and dental caries. However, advances in the field of genetic research combining new era of genetic engineering research may help clinicians to enhance treatment strategies and improve treatment outcomes to patients.<sup>29</sup> It is obvious that the salivary flow is hampered in the patients using beta-agonists as they have a negative effect on salivary production and many of the inhalers have low pH. In fact, due to dry mouth obviously, there is a thrust for intake of soft drinks, which could result in erosion of teeth. A similar sequence was found in the present case.

### TREATMENT PLAN

The composite resin restorations, the orientation of oral hygiene and the fabrication of overlay removable upper and lower dentures were accomplished in the present case.

### CONCLUSION

Developmental disorders such as AI can impact the quality of oral health and cause psychological effects on them. The observations in this case clearly indicate the association between enamel defects and systemic conditions such as asthma and dietary factors such as the intake of soft drinks.

However, an explanation for these associations requires molecular, functional, and genetic research integrating the inflammatory process of asthma and AI. Early diagnosis and implementation of dental treatment focusing on patient's needs with utmost care may help in preventing the progressive damage of teeth in future.

### REFERENCES

1. Simmer JP, Fincham AG. Molecular mechanisms of dental enamel formation. *Crit Rev Oral Biol Med.* 1995;6:84–108. <https://doi.org/10.1177/10454411950060020701>
2. Roma M, Hegde P, Durga Nandhini M, Hegde S. Management guidelines for amelogenesis imperfecta: A case report and review of the literature. *J Med Case Rep.* 2021;15:67–4. <https://doi.org/10.1186/s13256-020-02586-4>
3. Mauprivez C, Nguyen JF, de la Dure-Molla M, Naveau A. Prosthetic rehabilitation of a patient with rare and severe enamel renal syndrome. *Int J Prosthodont.* 2017;31:31–4. <https://doi.org/10.11607/ijp.5322>
4. Neville BW, Douglass DD, Allen CM, Bouquot JE. Abnormalities of teeth. In: *Oral and maxillofacial pathology.* Philadelphia, PA: Elsevier; 2004. p. 89–94.
5. Moreira RF, Figueiredo RG, Oliveira HE, da Fonseca ACL, de Miranda MS. Immediate desensitization in teeth affected by amelogenesis imperfecta. *Braz Dent J.* 2016;27:359–62. <https://doi.org/10.1590/0103-6440201600701>
6. Rodrigues VC, Sommer AA, Pereira RMA, Santana ACS, Andrade RSD. Amelogenesis imperfecta from diagnosis to rehabilitation – A case report. *Int J Case Rep Rev.* 2021;7(3):127. <https://doi.org/10.31579/2690-4861/127>

7. Wogelius P, Viuff JH, Haubek D. Use of asthma drugs and prevalence of molar incisor hypomineralization. *Int J Paediatr Dent.* 2020;30(6):734–40. <https://doi.org/10.1111/ipd.12655>
8. Khodaeian N, Sabouhi M, Ataei E. An Interdisciplinary approach for rehabilitating a patient with amelogenesis imperfecta: A case report. *Case Rep Dent.* 2012;2012:432108. <https://doi.org/10.1155/2012/432108>
9. Seymen F, Lee KE, Koruyucu M, Gencay K, Bayram M, Tuna EB, et al. Novel ITGB6 mutation in autosomal recessive amelogenesis imperfecta. *Oral Dis.* 2015;21:456–61. <https://doi.org/10.1111/odi.12303>
10. Poulsen S, Gjørup H, Haubek D, Haukali G, Hintze H, Løvschall H, et al. Amelogenesis imperfecta – A systematic literature review of associated dental and oro-facial abnormalities and their impact on patients. *Acta Odontol Scand.* 2008;66(4):193–9. <https://doi.org/10.1080/00016350802192071>
11. Gadhia K, McDonald S, Arkutu N, Malik K. Amelogenesis imperfecta: An introduction. *Br Dent J.* 2012;212:377–9. <https://doi.org/10.1038/sj.bdj.2012.314>
12. Aldred MJ, Savarirayan R, Crawford PJM. Amelogenesis imperfecta: A classification and catalogue for the 21st century. *Oral Dis.* 2003;9:19–23. <https://doi.org/10.1034/j.1601-0825.2003.00843.x>
13. Smith CE, Poulter JA, Antanaviciute A, Kirkham J, Brookes SJ, Inglehearn CF, et al. Amelogenesis imperfecta; genes, proteins, and pathways. *Front Physiol.* 2017;8:435. <https://doi.org/10.3389/fphys.2017.00435>
14. Shivhare P, Shankarnarayan L, Gupta A, Sushma P. Amelogenesis imperfecta: A review. *J Adv Oral Res.* 2016;7(1):1–3. <https://doi.org/10.1177/2229411220160101>
15. Chanmougananda SC, Ashokan KA, Ashokan SC, Bojan AB, Ganesh RM. Literature review of amelogenesis imperfecta with case report. *J Indian Acad Oral Med Radiol.* 2012;24:83–7. <https://doi.org/10.5005/jp-journals-10011-1266>
16. Guergolette RP, Cilene Dezan C, Garbelini Frossard WT, de Andrade Ferreira FB, Cerci Neto A, Fernandes KBP. Prevalence of developmental defects of enamel in children and adolescents with asthma. *J Bras Pneumol.* 2009;35(4):295–300. <https://doi.org/10.1590/S1806-37132009000400002>
17. Wogelius P, Haubek D, Nechifor A, Nørgaard M, Tvedebrink T, Poulsen S. Association between use of asthma drugs and prevalence of demarcated opacities in permanent first molars in 6-to-8-year-old Danish children. *Community Dent Oral Epidemiol.* 2010;38(2):145–51. <https://doi.org/10.1111/j.1600-0528.2009.00510.x>
18. Van Amerogen W, Kreulen C. Chesse molars: A pilot study of the etiology of hypocalcifications in first permanent molars. *J Dent Child.* 1995;62:266–9.
19. Tung K, Fujita H, Yamashita Y, Takagi Y. Effect of turpentine-induced fever during the enamel formation of rat incisor. *Arch Oral Biol.* 2006;51:464–70. <https://doi.org/10.1016/j.archoralbio.2005.12.001>
20. Tourino LFPG, Corrêa-Faria P, Ferreira RC, Bendo CB, Zarzar PM, Vale MP. Association between molar incisor hypomineralization in schoolchildren and both prenatal and postnatal factors: A population-based study. *PLoS One.* 2016;11(6):e0156332. <https://doi.org/10.1371/journal.pone.0156332>
21. Mastora A, Vadiakas G, Agouropoulos A, Gartagani-Panagiotopoulou P, Gemou Engesaeth V. Developmental defects of enamel in first permanent molars associated with use of asthma drugs in preschool aged children: A retrospective case-control study. *Eur Arch Paediatr Dent.* 2017;18:105–11. <https://doi.org/10.1007/s40368-017-0280-1>
22. Van Eygen I, Vannet BV, Wehrbein H. Influence of a soft drink with low pH on enamel surfaces: an in vitro study. *Am J Orthod Dentofacial Orthop.* 2005;128(3):372–7. <https://doi.org/10.1016/j.ajodo.2004.03.036>

23. Wang YL, Chang CC, Chi CW, Chang HH, Chiang YC, Chuang YC, et al. Erosive potential of soft drinks on human enamel: An in vitro study. *J Formos Med Assoc.* 2014;113(11):850–6. <https://doi.org/10.1016/j.jfma.2014.06.002>
24. Walsh LJ. Dry mouth: A clinical problem for children and young adults. *Minim Interv Dent.* 2009;2(1):55.
25. Temmel AF, Quint C, Schickinger-Fischer B, Hummel T. Taste function in xerostomia before and after treatment with a saliva substitute containing carboxy-methylcellulose. *J Otolaryngol.* 2005;34:116–20. <https://doi.org/10.2310/7070.2005.04008>
26. Young W, Khan F, Brandt R, Savage N, Razek AA, Huang Q. Syndromes with salivary dysfunction predispose to tooth wear: Case reports of congenital dysfunction of major salivary glands, Prader-Willi, congenital rubella, and Sjogren's syndromes. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2001;92(1):38–4. <https://doi.org/10.1067/moe.2001.113549>
27. Longman LP, McCracken CF, Higham SM, Field EA. The clinical assessment of oral dryness is a significant predictor of salivary gland hypofunction. *Oral Dis.* 2000;6(6):366–70. <https://doi.org/10.1111/j.1601-0825.2000.tb00128.x>
28. Koruyucu M, Kuvvetli SS, Tuna EB, Ozdas DO. Editorial: Amelogenesis imperfecta. *Front Dent Med.* 2022;3:888122. <https://doi.org/10.3389/fdmed.2022.888122>