CASE REPORT

Angioedema in a 9-year-old Child after Dental Treatment: A Rare Complication Explored through a Case Report

Katherina S Barman[1], Rena Ephraim[2], Prathima G Shivashankarappa[3], Anagha Chonat[4]

Abstract

Aim and background: Angioedema is a nonpruritic swelling that typically affects the skin, mucous membranes of the face, and perioral soft tissues. It can be life-threatening, but it is usually not and can be treated conservatively unless the airway is compromised. This paper seeks to illuminate a rare case of hereditary angioedema (HAE) onset following dental procedures in a 9-year-old Indian boy.

Case description: A 9-year-old male patient reported a chief complaint of spacing in the upper anterior region, which was diagnosed to be due to impacted supernumeraries. Two days after the oral surgical procedure, the child developed symptoms of periorbital edema with facial swelling. A second episode occurred a day after the delivery of the orthodontic appliance. This was also associated with facial swelling, respiratory distress, and gastrointestinal (GI) symptoms. A diagnosis of angioedema was confirmed and was treated appropriately.

Conclusion: Dental professionals must be aware of the possibility of triggering AE, a potentially fatal condition in patients. This case highlights the importance of comprehensive medical history intake and timely physician collaboration when confronting unexpected symptoms following a dental procedure.

Clinical significance: Awareness of rare conditions like HAE can aid dental professionals in early identification and appropriate management, preventing dangerous exacerbations and contributing to safer dental care.

Keywords: Angioedema, Case report, Dental procedures, Hereditary angioedema, Orthodontic appliance, Pediatric dentistry, Supernumerary teeth.

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Introduction

Hereditary angioedema (HAE) is a potentially fatal autosomal dominant disorder. The clinical appearance is characterized by recurring episodes of edema in the face, upper airway, gastrointestinal (GI) system, skin, and urethra.[1] The absence or malfunction of the C1 esterase inhibitor (C1-INH), a plasma protein that inhibits bradykinin activation and produces increased vascular permeability and edema, is the cause of this condition.[2] Several cases of HAE affecting adults have been reported from India.[3] However, no case of HAE affecting young children has been reported. Here, we present a case of angioedema in a 9-year-old child following dental treatment procedures.

Case Description

A 9-year-old male child came to the Department of Pedodontics and Preventive Dentistry with the primary complaint of upper front tooth spacing. A medical history review found that there was no medical or familial history of sickness. The orthopantomogram (OPG) showed two impacted supernumerary teeth between the upper two central incisors (Fig. 1). Following an investigation, it was determined that the best course of action would be to extract supernumerary teeth followed by orthodontic therapy. The supernumeraries were retrieved under local anesthesia with 2% lignocaine hydrochloride and a vasoconstrictor (Fig. 2). Resorbable vicryl rapide 2-0 sutures were used, and the parent and child received postextraction instructions. The patient was requested to return after a week for review. Only 3 months later, the parent and their child reported to the clinic.

When asked about the delay in reporting for review, the parent stated that the child had developed symptoms of periorbital swelling followed by facial swelling 2 days after extraction. They sought emergency treatment at the local Government General Hospital. There was no prior bug bite, eating of new food, or contact

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Patient consent statement: The author(s) have obtained written informed consent from the patient's parents/legal guardians for publication of the case report details and related images.

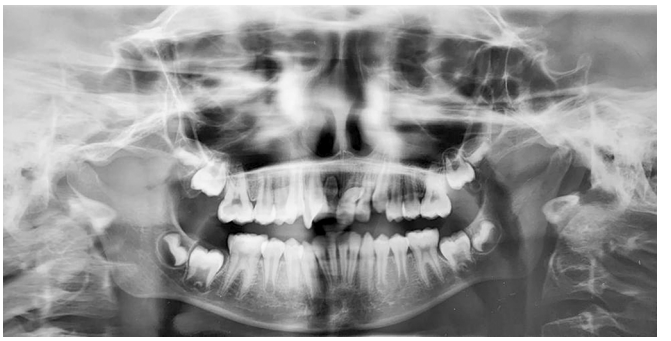

Fig. 1: OPG revealing two supernumeraries between 11 and 21

with latex. The hospital's on duty emergency doctor ordered systemic antihistamines. According to the parent, the symptoms did not improve significantly but faded on their own over time. This was the child's first case of swelling and his first visit to the dentist.

On intraoral examination, the extraction site had healed well, and the child was medically fit for future orthodontic treatment, as anticipated (Fig. 3). A 2 × 4 appliance was provided, and the patient was given postoperative instructions to follow a soft diet and use mouthwash (Figs 4 and 5). The following day, the child and parent returned to the department with facial swelling, difficulty breathing, and GI symptoms of diarrhea. His physical examination revealed periorbital edema with submental fullness, but no papular skin rash, including the face, trunk, or upper limbs. There were no clinical signs of urticaria. The child was sent to a medical facility for emergency treatment and further examination. Because the boy had previously not responded to antihistamines, he was treated like any other angioedema attack, using epinephrine to diminish edema and salbutamol to relieve airway symptoms.

Once the symptoms had abated, blood tests for hematologic analysis were performed. The C4, C-INH, and C1-INH levels were all low. The clinical diagnosis of HAE was established. After a week, the child's symptoms abated, and when discharged from the hospital, the parent indicated her unwillingness to undergo further dental treatment, thus the orthodontic appliance was debonded.

Discussion

Evidence linking dental procedures to HAE exacerbation in adults has been well-documented in the literature.[4–7] However, case reports in pediatric populations remain scarce.[8] It is crucial for pediatric dentists to be cognizant of HAE, as routine dental interventions may precipitate life-threatening complications such as laryngeal edema, airway obstruction, or hypoxia.[9,10] These adverse events can manifest hours or even days after the triggering procedure, long after the dental appointment has concluded.[11,12] In the present case, the dental intervention was performed in the early afternoon, with swelling manifesting 2 days later in the morning. Consequently, dental practitioners may be unaware of the patient's deteriorating condition posttreatment and unable to provide timely assistance during a critical period.

The European Academy of Allergy and Clinical Immunology has delineated four categories of acquired angioedema, alongside three distinct types of HAE, all of which follow an autosomal dominant inheritance pattern. In HAE types 1 and 2, symptom onset can occur as early as 2 years of age, with a mean onset age of 8–12 years, and edematous episodes typically persist for 2–5 days. HAE type 3 exhibits a later mean age of symptom onset at 27 ± 14 years. With the exception of the exceedingly rare HAE type 3, which demonstrates a marked female predominance, HAE exhibits no significant ethnic or gender predilection.[13]

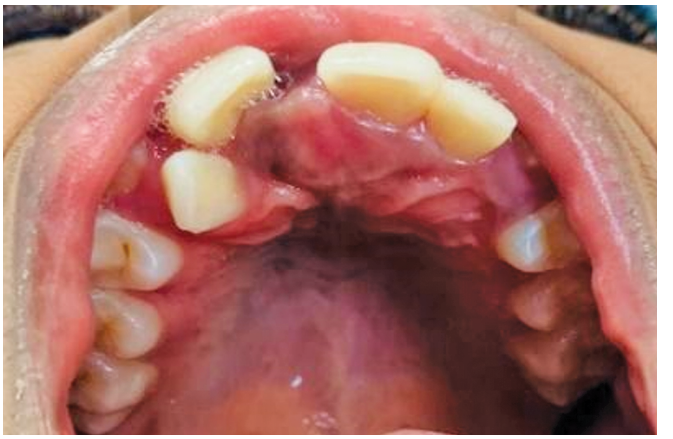

Fig. 3: Re-epithelialization and healed alveolar bone socket

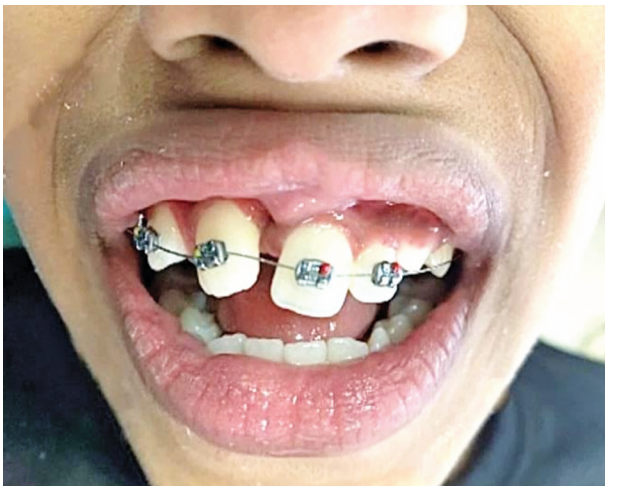

Fig. 4: 2 × 4 appliance in place

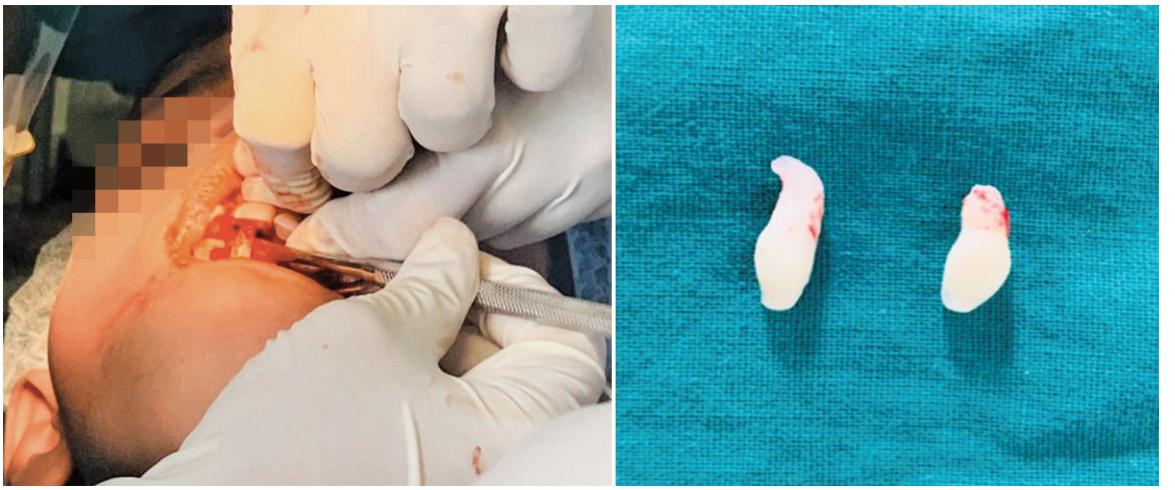

Fig. 2: Extraction of supernumeraries

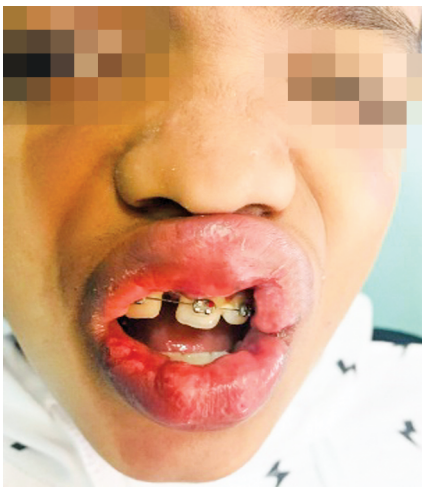

Fig. 5: Periorals edema with facial swelling

HAE is characterized by recurrent nonpitting edema, predominantly affecting the skin and GI tract. The affected skin typically presents as swollen, tender, and warm, but notably without accompanying urticaria.[14] While a burning sensation is frequently reported, pruritus is uncommon.[15] Episodes generally persist for 48–72 hours, though they may extend up to 1–2 weeks. The onset of an acute attack is often associated with stress, trauma, infection, or temperature fluctuations.[16] This etiology elucidates the edematous episodes observed in our case following both the extraction procedure and the placement of the orthodontic appliance (2 × 4 appliance). Our patient's presentation included facial swelling without urticaria or pruritus, accompanied by GI manifestations.

The angioedema episodes were of moderate severity, accompanied by respiratory and GI symptoms. Given the clinical presentation of perioral edema and facial swelling in the absence of urticaria or pruritic rashes, a provisional diagnosis of HAE was established. It is noteworthy that the patient's parent may be unaware of similar experiences in other family members. Of particular concern to dental practitioners is the potential for dental interventions to precipitate HAE episodes. Van Sickels et al. have reported that procedures as routine as dental impression-taking or pulpal excavation can trigger potentially life-threatening laryngeal edema, typically developing 24–48 hours posttreatment, potentially resulting in airway obstruction and mortality. However, it is important to note that not every dental extraction necessarily leads to an acute episode.[17] HAE attacks can be provoked by both physical and psychological stressors, and dental appointments present numerous potential triggers.[16]

It is also possible that the local anesthetic administered in this case influenced the outcome.[18] If the patient has been diagnosed with HAE at the time of presentation, management should be done in consultation with their physician.[11] Although HAE can be treated medically, they specialize in acute attacks. If HAE is suspected, it is appropriate to consult with the patient's doctor to explore treatment options.

Conclusion

In conclusion, we concur that every dental professional ought to be aware of the disorder known as HAE. Although there is a little chance of running into one of these patients, the effects of uninformed care could be very bad. We advise dental professionals to add a question to their dental history forms that asks about prior recurrent episodes of facial swelling and breathing problems as a precaution to prevent more tragic consequences. As an alternative, practitioners could be aware of the situation while asking patients about their general and medical histories so that they can identify any cues that may be disclosed and respond appropriately.

Clinical Significance

This case underscores the importance of awareness among dental professionals about HAE, especially since dental procedures can trigger potentially fatal HAE episodes. The case highlights the need for thorough medical history taking, swift recognition of symptoms, and prompt referral for appropriate medical intervention. It advocates for interdisciplinary collaboration for optimal patient management. Furthermore, it emphasizes the critical role of informed consent and patient education in preventing severe episodes. Lastly, this case suggests the need for continued research on HAE triggers and management, particularly in pediatric patients and in the context of dental procedures.

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References

1. Azmy V, Brooks JP, Hsu FI. Clinical presentation of hereditary angioedema. Allergy Asthma Proc 2020:41:S18–S21.
2. Agostoni A, Aygoren-Pursun E, Binkley KE, et al. Hereditary and acquired angioedema: problems and progress: proceedings of the third C1 esterase inhibitor deficiency workshop and beyond. J Allergy Clin Immunol 2004;114:S51–S131.
3. Jindal AK, Bishnoi A, Dogra S. Hereditary angioedema: diagnostic algorithm and current treatment concepts. Indian Dermatol Online J 2021;12(6):796.
4. Heft MW, Flynn PM. Hereditary angioedema: review of literature and dental treatment. J Am D Assoc 1977;95(5):986–990.
5. Lee YS, Chung JH, Cho KH, et al. A case of hereditary angioedema. Korean J Dermatol 1994;32:115–118.
6. Suh KS, Kang JM, Kim KJ, et al. Three cases of hereditary angioedema in one family. Korean J Dermatol 1995;33:564–569.
7. Lee JA, Nah BG, Jun H, et al. A case of hereditary angioedema not manifestated classical autosomal dominant trait. Korean J Allergy 1997;17:574–579.
8. Shin M, Ahn K. A case of hereditary angioedema in a 7-year-old Korean girl. Allergy Asthma Immunol Res 2013;5(1):59–61.
9. Rosa A, Miranda M, Franco R, et al. Experimental protocol of dental procedures in patients with hereditary angioedema: the role of anxiety and the use of nitrogen oxide. Oral Implantol 2016;9(2):49.
10. Bork K, Hardt J, Schicketanz KH, et al. Clinical studies of sudden upper airway obstruction in patients with hereditary angioedema due to C1 esterase inhibitor deficiency. Arch Int Med 2003;163:1229–1235.
11. Bork K. Recurrent angioedema and the threat of asphyxiation. Dtsch Arztebl Int 2010;107(23):408.
12. Morcavallo PS, Leonida A, Rossi G, et al. Hereditary angioedema in oral surgery: overview of the clinical picture and report of a case. OMFS 2010;68(9):2307–2311.
13. Zuberbier T, Abdul Latiff AH, Abuzakouk M, et al. The international EAACI/GA²LEN/EuroGuiDerm/APAACI guideline for the definition, classification, diagnosis, and management of urticaria. Allergy 2022;77(3):734–766.
14. Carreer FM. The C1 inhibitor deficiency. A review. Eur J Clin Chem Clin Biochem 1992;30:793–807.

15. Bork K, Meng G, Staubach P, et al. Hereditary angioedema: new findings concerning symptoms, affected organs, and course. Am J Med 2006;119(3):267–274.
16. Sanuki T, Watanabe T, Kurata S, et al. Perioperative management of tooth extractions for a patient with hereditary angioedema. J Oral Maxillofac Surg 2014;72:2421.e1–2421.e3.
17. Van Sickels NJ, Hunsaker RB, Van Sickels JE. Hereditary angioedema: treatment, management, and precautions in patients presenting for dental care. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;109:168–172.
18. Septodont. Scandonest.pdf. Available at http://www.septodont.in/sites/default/files/Scandonest.pdf. Accessed on 16 May 2016.