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## HAMAMY SENDROMLU BİR HASTANIN ENDODONTİK YENİDEN TEDAVİSİ: BİR OLGU SUNUMU



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### ÖZET

Enfeksiyon nedeniyle apikal bariyerini kaybetmiş dişler için mineral trioksit agregat ile apikal tıkaç oluşturmak faydalıdır. Bu vaka raporu, Hamamy Sendromlu bir hastada eksternal kök rezorpsiyonu bulunan sol mandibular birinci premolar dişin endodontik yeniden tedavisini sunmayı amaçlamaktadır. 21 yaşında hamamy sendromu olan erkek hasta, mandibula sol birinci premolarında spontan ağrı ve şişlik şikayeti ile sevk edildi. Diş akut apikal apse teşhisi kondu. İlgili diş daha önce bölümümüzdeki bir endodonti uzmanı tarafından yenilenmişti. Diş yapısı nedeniyle apikal cerrahi uygulanamadı. Hastaya endodontik yeniden tedavi planlandı.

**Anahtar Kelimeler:** *Eksternal Kök Rezorpsiyonu, Endodontik Yeniden Tedavi, Hamamy Sendromu*

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## ENDODONTIC NON-SURGICAL RETREATMENT OF PATIENT WITH HAMAMY SYNDROME: A CASE REPORT

### Summary

Creating apical plug formation with mineral trioxide aggregate (MTA) is beneficial for teeth that have lost their apical barrier due to infection. This case report aims to present the retreatment of a left mandibular first premolar tooth with external root resorption of a patient with hamamy syndrome. A 21-year-old male patient suffering from hamamy syndrome was referred complaining of spontaneous pain and swelling in his mandibular left first premolar. The tooth was diagnosed with an acute apical abscess. It was previously retreated by an endodontic specialist in our department. Apical surgery was not applicable because of the tooth structure and second endodontic retreatment was performed.

**Keywords:** *External Root Resorption, Endodontic Retreatment, Hamamy Syndrome*

### Introduction

Craniofacial dysplasia-osteopenia syndrome, also known as Hamamy Syndrome (HS)<sup>1</sup>, is a quite uncommon disease. Mutation of IRX5 gene on chromosome 16q12.2. causes this

disease<sup>2</sup>.

This syndrome has defined by Hamamy et al. and his patients were brothers born to double first-cousin and had severe hypertelorism, upslanting palpebral fissures, brachycephaly, atypical ears, sloping shoulders, enamel hypoplasia, and osteopenia with bone fractures. Both had severe myopia, mild to moderate sensorineural hearing loss, and borderline intelligence. The oral findings of his patients were; wide mouth, thin upper vermilion border, flat philtrum, mild micrognathia, highly arched palate, thin enamel, and loss of lamina dura<sup>3</sup>. After years of the author's first presentation now it recorded that the patients who have this syndrome suffer from developmental delay, intellectual disability, severe telecanthus, abnormal ears, dentinogenesis imperfecta, bone fragility<sup>4</sup>, anemia, and congenital heart defects<sup>2</sup>.

External resorption is pathological in permanent dentition and, it can not be reversed. Because of its aggressive and progressive character, it causes damage to tooth hard tissue which can lead to early loss of affected teeth. In apically damaged

teeth because of external resorption, one of the issues is to obtain a well-sealed apical area <sup>5</sup>. In addition to professional experience and education and of course sensible case selection, materials can play an important role in such cases.

After its approval by the U.S. Food and Drug Administration <sup>6</sup>, Mineral Trioxide Aggregate (MTA) cement has become very useful in many cases in the endodontic area, including root canal filling <sup>7,8</sup> and, it can be used as not only retrograde but also orthograde filling material for both open or closed apices <sup>9,10,11,12</sup>. With its usage of plug formation aiming to create a proper seal in apically deformed teeth <sup>13,14,15</sup>, we benefit from its effect on the osteogenesis process <sup>16</sup> which is related to its content. MTA includes bioactive elements, and via this, it can be a repair tool

for damaged tissues surrounding teeth <sup>17</sup>.

In this case report, we aim to present the retreatment of a left mandibular first premolar tooth with external root resorption in a patient who has HS.

### Case Report

A 21-year-old male patient was referred to Inonu University Faculty of Dentistry, Department of Endodontics complaining of spontaneous pain and swelling in his mandibular left first premolar. With medical history, the patient's HS and the presence of osteogenesis imperfecta disorder were learned. The patient we had presented in this case report was previously mentioned by other researchers <sup>18</sup>.

The radiographic examination

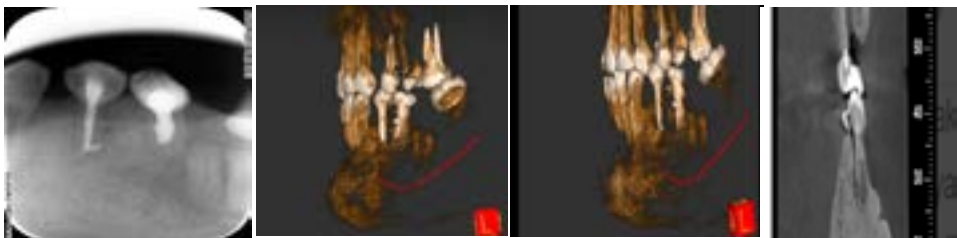
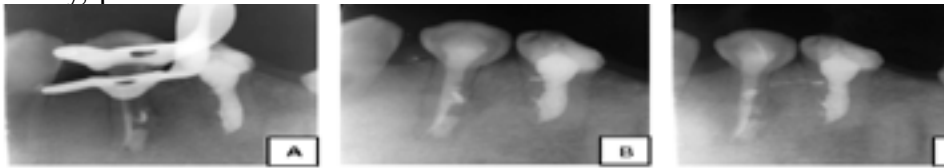


Figure 1 Before Treatment

showed an apical lesion and resorption areas in the apical third of the root (Figure 1).

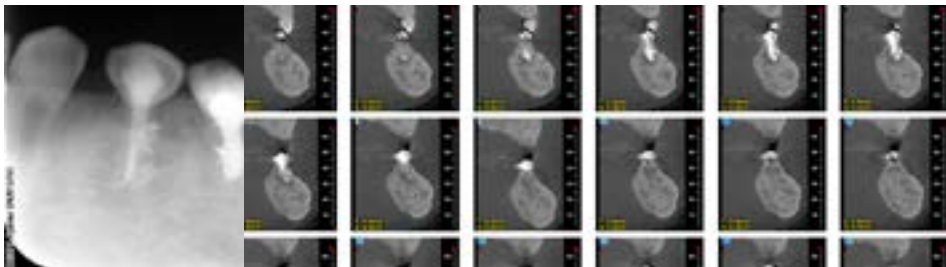
The tooth had a single root and a single canal and was diagnosed with an acute apical abscess. The tooth was previously retreated by an endodontic specialist in our department. Apical surgery was not applicable because of the tooth structure. It was decided to perform retreatment of the tooth with the consent of the patient. Before retreatment cone beam computed tomography images were obtained. After the preparation of the access cavity, previous root canal obtura-

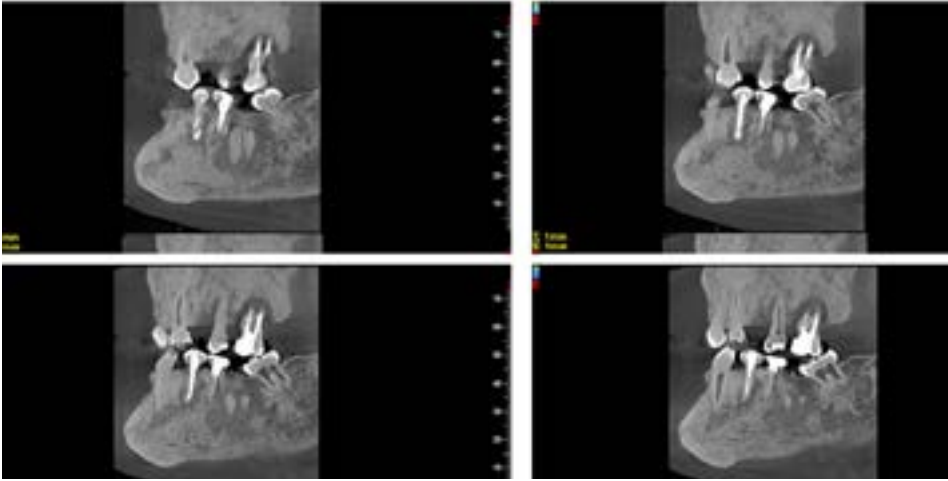
tion materials were removed with hand files and R Endo Retreatment Files (MICRO-MEGA). The working length was detected with an apex locator (VDW.GOLD RECIPROC Endo motor with integrated apex locator) and verified with periapical radiography. The root canal was cleaned and shaped with the crown down technique using 2.5% sodium hypochlorite and 17% EDTA irrigation. At the second appointment, the tooth was without symptoms, and the root canal was dressed with calcium hydroxide. 10 days later the root canal was completely obturated with MTA (ProRoot, Dentsply / Tul-



**Figure 2** A: During retreatment, B: After Permanent Restoration, C: After 6 Months Follow-up

In three years of follow-up, the tooth was asymptomatic and functional (Figure 3).





sa Dental Specialties). The composite restoration was applied the next following appointment (Figure 2).

Figure 3 After 3 Years Follow-up

### Conclusion

The hamamy syndrome is a very rare condition. It affects the entire body as well as orofacial tissues. Patients who have these types of syndromes can refer to a dentist to seek for treatment which may be difficult to treat even for patients with good systemic health. Root resorption is one

of these dental diseases. In our case, root resorption highly occurred due to persistent infection. However, the syndrome of the patient involving bone and tooth disorder might keep the potential to affect the dental resorption progression and prognosis. The other issue is the procedures of retreatment which might easily cause more fragile dentin walls and the irregularly shaped apical structure. Providing optimum standards for endodontic treatment is a concern for both practitioner and patient. Developments in dental materials whi-

ch increase the potential of healing of teeth and surrounding tissues are important for providing better solutions for resorption treatment.

## References

1. Buget, M. I., & Kucukay, S. (2021). Hamamy syndrome. *AnästH Intensivmed*, 62, S191-S196.
2. Bonnard, C., Strobl, A. C., Shboul, M., Lee, H., Merriman, B., Nelson, S. F., ... & Reversade, B. (2012). Mutations in IRX5 impair craniofacial development and germ cell migration via SDF1. *Nature genetics*, 44(6), 709-713.
3. Hamamy, H. A., Teebi, A. S., Oudjhane, K., Shegem, N. N., & Ajlouni, K. M. (2007). Severe hypertelorism, midface prominence, prominent/simple ears, severe myopia, borderline intelligence, and bone fragility in two brothers: new syndrome?. *American Journal of Medical Genetics Part A*, 143(3), 229-234.
4. Mégarbané, A., Hana, S., Mégarbané, H., Castro, C., Baulande, S., Criqui, A., ... & Delague, V. (2021). Clinical and Molecular Update on the Fourth Reported Family with Hamamy Syndrome. *Molecular Syndromology*, 12(6), 342-350
5. Patel, S., Saberi, N., Pimental, T., & Teng, P. H. (2022). Present status and future directions: Root resorption. *International Endodontic Journal*.
6. Schwartz, R. S., Mauger, M., Clement, D. J., & WALKER III, W. A. (1999). Mineral trioxide aggregate: a new material for endodontics. *The Journal of the American Dental Association*, 130(7), 967-975.
7. An, H. J., Yoon, H., Jung, H. I., Shin, D. H., & Song, M. (2021). Comparison of obturation quality after MTA orthograde filling with various obturation techniques. *Journal of Clinical Medicine*, 10(8), 1719.
8. Siboni, F., Taddei, P., Prati, C., & Gandolfi, M. G. (2017). Properties of Neo MTA Plus and MTA Plus cements for endodontics. *International endodontic journal*, 50, e83-e94.
9. Torabinejad, M., Parirokh, M., & Dummer, P. M. (2018). Mineral trioxide aggregate and other bioactive endodontic cements: an updated overview—part II: other clinical applications and complications. *International endodontic journal*, 51(3), 284-317.
10. Karunakar, P., Rangareddy,

- M. S., Karteek, B., Lakshmi, C., & Reddy, C. Management OfTeeth With Open Apices Using MTA As Orthograde Filling Material–A Case Report.
11. Mohammadi, Z. (2008). Sealing ability of MTA cements as orthograde root filling materials. *Pesquisa Brasileira em Odontopediatria e Clínica Integrada*, 8(3), 267-270.
12. Keleş, A., Torabinejad, M., Keskin, C., Sah, D., Uzun, İ., & Alçin, H. (2018). Micro-CT evaluation of voids using two root filling techniques in the placement of MTA in mesial root canals of Vertucci type II configuration. *Clinical oral investigations*, 22(5), 1907-1913.
13. Cehreli, Z. C., Sara, S., Uysal, S., & Turgut, M. D. (2011). MTA apical plugs in the treatment of traumatized immature teeth with large periapical lesions. *Dental Traumatology*, 27(1), 59-62.
14. Floratos, S. G., Tsatsoulis, I. N., & Kontakiotis, E. G. (2013). Apical barrier formation after incomplete orthograde MTA apical plug placement in teeth with open apex-report of two cases. *Brazilian Dental Journal*, 24, 163-166.
15. Pace, R., Giuliani, V., Pini Prato, L., Baccetti, T., & Pagavino, G. (2007). Apical plug technique using mineral trioxide aggregate: results from a case series. *International endodontic journal*, 40(6), 478-484.
16. Cervino, G., Laino, L., D'Amico, C., Russo, D., Nucci, L., Amoroso, G., ... & Fiorillo, L. (2020). Mineral trioxide aggregate applications in endodontics: A review. *European journal of dentistry*, 14(04), 683-691.
17. Bogen, G., & Kuttler, S. (2009). Mineral trioxide aggregate obturation: a review and case series. *Journal of endodontics*, 35(6), 777-790.
18. Duman, S. B., Dedeoglu, N., Arıkan, B., & Altun, O. (2020). Sphenoid sinus agenesis and sella turcica hypoplasia: very rare cases of two brothers with Hamamy syndrome. *Surgical and Radiologic Anatomy*, 42(11), 1377-1380