



A Unique Clinical Appearance of Upper Maxillary Premolar associated with Dentigerous Cyst: Case Report

Mohd Faizal Abdullah^{1,2*}, Shaifulizan Abdul Rahman^{1,2}, Nik Rozainah Nik Abdul Ghani³, Rosnani Mamat³, Masitah Hayati Harun⁴

¹Oral and Maxillofacial Surgery Unit, School of Dental Sciences, Universiti Sains Malaysia, Health Campus, 16150, Kubang Kerian, Kota Bharu, Kelantan

²Oral and Maxillofacial Surgery Unit, Hospital Universiti Sains Malaysia, Universiti, Sains Malaysia, Health Campus, 16150, Kubang Kerian, Kota Bharu, Kelantan.

³Conservative Unit, School of Dental Sciences, Universiti Sains Malaysia, Health Campus, 16150, Kubang Kerian, Kota Bharu, Kelantan.

⁴Oral Medicine and Oral Pathology Unit, School of Dental Sciences, Universiti Sains Malaysia, Health Campus, 16150, Kubang Kerian, Kota Bharu, Kelantan

ARTICLE INFO

Article history:

Received 6 April 2023

Revised 20 June 2023

Accepted 20 July 2023

Online first

Published 1st March 2024

Keywords:

dental anomaly
double-headed upper premolar
hypercementosis
dentigerous cyst

DOI:

10.24191/cos.v11i1.26043

ABSTRACT

During dental development, a variety of hereditary and environmental variables interact to cause dental abnormalities. The etiology may involve multi-tiered, multi-directional, and multi-factorial processes. Fusion is a dental anomaly that happens during tooth development, where two separated teeth bud fuse together to form a single tooth. The morpho-differentiation or histo-differentiation stages of tooth development, genetic and environmental variables can induce developmental dental abnormalities. A clinical resemblance of a "double-headed" maxillary second upper left premolar with two crowns, formed on the both coronal and apical root end or whether it is just a hypercementosis of apical root discovered after surgical extraction and enucleation of cyst. Investigations to confirm such unique findings were performed. This case report discusses the importance of clinical, radiographic and laboratory investigations to confirm the unique appearance of the maxillary second premolar

INTRODUCTION

The term "double tooth" is widely used to denote a conjoined set of teeth. It is possible that this is related to the fusion of two distinct tooth buds (Hunasgi et al. 2017). Union may be full or imperfect, and the tooth's root canals may be separate or fused, depending on the stage of development. The condition is more common in deciduous than in permanent dentition. Dental anomalies can lead to significant functional as well as aesthetic issues in both jaws (Fekonja 2017). There were 11 reported dental anomalies and these anomalies were divided based on five categories (number, size, shape, position and eruption)(Sella Tunis et al. 2021): Fusion can occur between teeth of the same dentition or mixed dentitions. Commonly, a fused

^{1*} Corresponding author. E-mail address: mohdfaizalkk@usm.my

tooth is asymptomatic (Nunes et al. 2002). Supernumerary teeth are frequently atypical and have a cone-like clinical appearance. As a result, when a supernumerary to normal tooth is fused, the two portions of the united crown will usually display variations.

Understanding the morphogenesis process and the variability in outcomes is critical to the multidisciplinary clinical team's treatment approach. Early detection provides better patient care and treatment planning, as well as a reduction in complications and complexity of treatment. If such genetic correlations exist, they may be worth recognising and investigating, as early detection of developmental disturbance may disclose a possible risk of eruption disorders. In clinical practice, multiple dental abnormalities of the dentition are regularly found (Patil et al. 2013).

The organisation of the dental arches can be affected by changes in tooth eruption patterns, resulting in a malocclusion. The hereditary component of the reported tooth disruption can be investigated from a clinical standpoint by examining the associated dental malformations.

Hypercementosis is characterised by the excessive deposition of non-neoplastic cementum over normal root cementum, which alters root morphology. This cementum can be either hypocellular or cellular. The aetiology of hypercementosis is unknown (Shoor et al. 2014).

CASE DESCRIPTION

A 63-year-old man presented to endodontic clinic with non-resolving swelling in the upper left premolar region associated with intermittent pus discharge. Patient had a history of root canal treatment and porcelain-bonded metal crown 10 years ago. He reported his tooth 24 is congenitally missing. An intraoral periapical radiograph revealed a poorly obturated canal of tooth 25 and a well-defined radiolucency surrounding the bulbous root. Cone beam computed tomography (CBCT) revealed another crown-like structure resembling an upper premolar crown at the root apex of tooth 25. He was referred to Oral and Maxillofacial clinic for further treatment. Cyst enucleation and extraction of tooth 25 was performed under general anaesthesia.

The tooth specimen had two crowns, one on the coronal part and one at the apex, respectively. Tooth 25 was capped with porcelain fused to a metal crown, and has a single root ending. On the apical side, a less well-developed crown having a closer anatomic morphology to that of the maxillary premolar crown was observed. "Crown" on the apical end showed incomplete formation that surrounds the root apex of 25. The cystic lining was seen enveloping the less well developed "crown" at the apical end of 25. Histopathological examination (HPE) revealed a lesion consistent with an inflamed odontogenic cyst. Figure 1(A-F) depicts clinical, radiographic, operative, and photomicrograph of the H&E-stained cystic tissue.

Due to the unique presentation of the double-headed premolar, we conducted a three-dimensional (3D) study of this tooth using MIMIC software version 21.0. The complexity of the root canal system of this tooth was appreciated in this study with the presence of multiple root canal systems, and incomplete apical and defect of coronal seal. The Hounsfield Unit (HU) for "crown" at the apical part was 2226 and was comparable with the mean HU unit of enamel (2050.75HU) based on the CBCT study (Marzook et al. 2010). The three dimensional images generation (Axial, Sagittal, and Coronal) and (HU) of the apical "crown" is shown in Figure 2(A-E). The clinical and histopathological findings supported the diagnosis of an infected dentigerous cyst associated with a "double headed premolar". Due to its rare phenomenon, sectioning of the so called "double-headed premolar" was done and ground sectioning was performed to

confirm the final verdict of exact structure at the apical end whether it is pure “enamel” or just a mere hypercementosis. The ground sectioning shown the presence of cementocytes which confirmed the lesion is hypercementosis. The sectioning of specimen, ground sectioning images were shown in Figure 3(A-D).

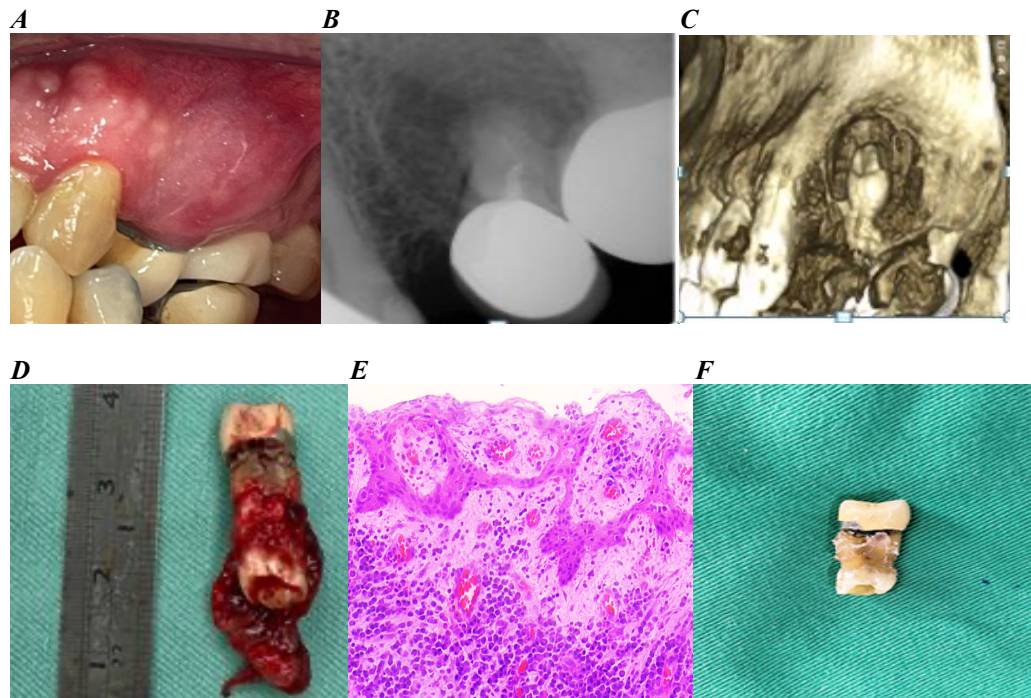


Fig. 1. A-F: Swelling at the buccal of 25, Bulbous root of 25 on intraoral periapical radiograph with insufficient obturation, CBCT showed a crown like structure, enucleated cystic lining with “double-headed premolar”, photomicrograph of inflamed odontogenic cyst tissue showing arcading lining epithelium (H&E, original magnification x400), and clinical appearance of double headed premolar.

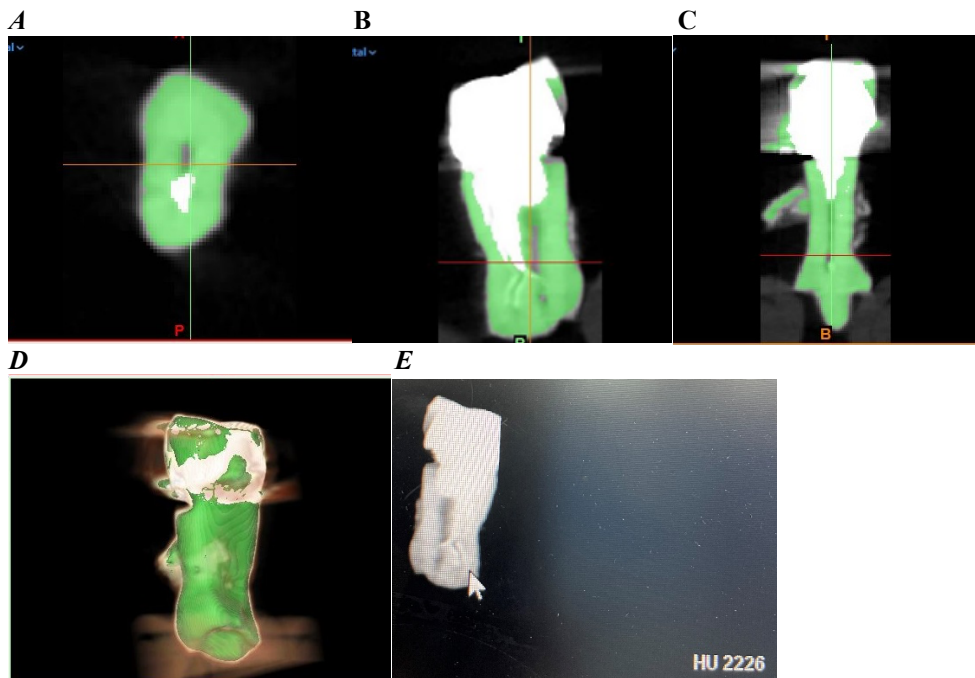


Fig. 2. A-E: Three- Dimensional (3D) images shown incomplete coronal seal and apical seal due to complexity of root canal system and respective HU measurement as shown by small arrow.

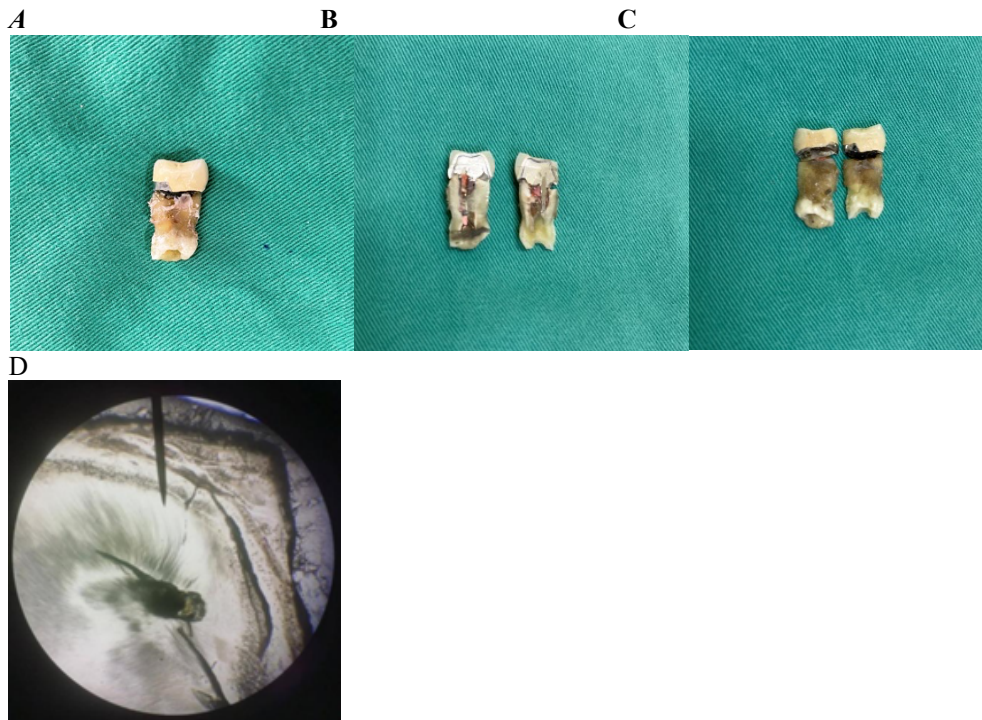


Fig. 3. A-D: Original clinical appearance of "Double Headed Premolar", Inner aspect of sectioned specimen, Outer aspect of sectioned specimen, Ground section of the specimen.

DISCUSSION

The aetiology of fusion is still debated, and several theories have been proposed (Fekonja 2017; Nunes et al. 2002; Patil et al. 2013; Sella Tunis et al. 2021). Epithelial and mesenchymal germ layers' fusion often results in aberrant tooth morphology (Nunes et al. 2002). Double tooth are two independent teeth that are joined by their dentin and (perhaps) their pulp. The tooth in our case exhibited two heads on either ends of the tooth. The authors hypothesized that the union may be due to the fusion of two separate tooth buds, but an entity such as an inverted supernumerary tooth cannot be overlooked (Hundal et al. 2016). The authors strongly believe this phenomenon is due to the fusion of the two separate tooth buds, given the clinical history of a congenitally missing first left premolar obtained from the patient.

This unusual location of the second tooth bud lying exactly beneath the first premolar during development may have resulted in this one-of-a-kind and extremely rare anomaly of a double-headed premolar in maxillary arch. These anomalies can result in treatment failure or periapical pathologies. One reported case of the double headed premolar in mandibular premolar was described by the author but were unable to trace the patient after extraction and hypothesized that pulpo-periodontal pathology was the primary reason for extraction (Sivakumar, Nair, and Raman 2010). The causes of endodontic treatment failure was extensively discussed (Tabassum and Khan 2016), and in this case, untreated both major and accessory canals, as well as improper coronal seal manifested in 3D study, were the causes of endodontic treatment failure. In our case, the previous dentist had missed a proper investigation and inspection, thus lead to a failure of the endodontic treatment.

In view of the inability to compare the HU for both end of the crowns due to coronal "crown" being lost due to previous porcelain bonded metal crown preparation, CBCT study of HU units of teeth (Marzook et al. 2010) was used and the apical "crown" HU unit was comparable to mean HU for enamel based on the above mentioned study.

The authors believed that such a tooth morphological anomaly could be caused by one or two factors. The first factor is the fusion of the first and second maxillary premolars from the same arch and second factor that can lead to this rare phenomenon is the fusion between premolar and supernumerary tooth. It is extremely rare for the second tooth bud to get fused right at the apical end of another tooth, but it was speculated in our case up to these stages of clinical and radiographic investigations. Even though speculative hypothesis of double headed maxillary premolar was made, due to extreme rarity of this phenomenon, confirmatory diagnosis is mandatory. Ground section showed presence of cementocytes instead of enamel structure confirmed the diagnosis of hypercementosis. Clinical, radiographic findings and HU alone are insufficient to diagnose a rare anomaly of so called "Double Headed Premolar".

Hypercementosis is typically associated with one or more local or systemic causes. The prevalence of hypercementosis by race or population group has yet to be determined. Mandibular molars are the most commonly affected teeth, followed by mandibular and maxillary second premolars and mandibular first premolars (Consolaro, Consolaro, and Francischone 2012). However, according to some authors, premolars are the most commonly affected teeth as in manifested in our case (Leider and Garbarino 1987b; Napier Souza et al. 2004).

Occlusal trauma, inflammation secondary to pulpal or periodontal disease, tooth mobility, root fracture repair, and tooth transplantation have all been linked to local causes of hypercementosis. We postulate that chronic inflammation secondary to pulpal disease and insufficient endodontic treatment are the causes that lead to hypercementosis of root end. A dentigerous cyst is most frequently found in individuals in the age

group between 20 and 40 years (Ochsenius et al. 2007). The mandibular third molars, maxillary third molars, maxillary canines, and premolars of both the maxillary and mandibular bones are where the majority of typical dentigerous cysts are found (Lustmann and Bodner 1988). Again we postulate that residual infection at the periapical region will lead to the hypercementosis and dentigerous cyst infection. Acromegaly, goitre, arthritis, rheumatic fever, calcinosis, Gardner's syndrome, Paget's disease, and vitamin A deficiency are all systemic conditions associated with hypercementosis (Manson-Hing 2005; Langlais, Langland, and Nortjé 1995; Leider and Garbarino 1987a).

The introduction of CBCT in the field of endodontics and maxillofacial surgery has resulted in a paradigm shift in clinical practice because it provides comprehensive preoperative and post-operative assessment of tooth morphology, existing periapical pathology, root resorption and monitor healing of periapical region post treatment (Scarfe et al. 2009).

CONCLUSION

Authors would like to conclude that even though with history of congenital missing maxillary left first premolar, clinical appearance of “apical” crown resembling an enamel structure, a CBCT appearance and HU of apical crown close to enamel structure reading, suggested of rare phenomenon of “Double Headed Premolar”, speculative clinical diagnosis should not be made until confirmatory test such as ground section was conducted. CBCT provides comprehensive assessment and useful for treatment planning.

Take home messages

- Proper investigations need to be requested to prevent inadequate treatment.
- Clinical, radiographic and software assessment not sufficient to conclude the rare entity of double headed premolar.
- Inadequate treatment will jeopardize patient oral health status.

CONFLICTS OF INTEREST

The authors declare no conflict of interest.

AUTHORS' CONTRIBUTIONS

MFA, the corresponding author and first author, conceived and provided the data for the case report and submitted the revised manuscript. SAR, the second author, collected and organized the data. NRNAG and RM, a co-author, provided logistic support as well as references collections, and MHH, a co-author, provided histopathological report and revise the manuscript. All authors have critically reviewed the case report and are responsible for the content and the manuscript.

ETHICAL APPROVAL

The patient's anonymity is carefully protected. We have obtained written consent of the patient and for the use of clinical information and photos for publication.

FUNDING

This case report does not receive any funding.

REFERENCES

- Consolaro, A., Consolaro, R. B., & Francischone, L. A. (2012). Cementum, apical morphology and hypercementosis: a probable adaptive response of the periodontal support tissues and potential orthodontic implications. *Dental Press Journal of Orthodontics*, 17(1), 21-30. <https://doi.org/10.1590/S2176-94512012000100003>.
- Fekonja, A. (2017). Prevalence of dental developmental anomalies of permanent teeth in children and their influence on esthetics. *Journal of Esthetic and Restorative Dentistry*, 29(4), 276-83. <https://doi.org/10.1111/jerd.12302>.
- Hunasgi, S., Koneru, A., Manvikar, V., Vanishree, M., & Amrutha, R. (2017). A Rare Case of Twinning Involving Primary Maxillary Lateral Incisor with Review of Literature. *Journal of Clinical Diagnostic Research*, 11(2), ZD09-ZD11. <https://doi.org/10.7860/JCDR/2017/23510.9212>.
- Hundal, K., Kaur, R., Sharma, S., Banik, S., & Arora, K. S. (2016). Occurrence of double mesiodens and an inverted mesiodens: a report of 2 rare cases. *Journal of Oral Medicine, Oral Surgery, Oral Pathology and Oral Radiology*, 2(3), 187-89. <https://doi.org/10.5958/2395-6194.2016.00044.8>.
- Langlais, R. P., Langland, O. E., & Nortjé, C. J. (1995). Diagnostic imaging of the jaws. Lea & Febiger (Eds.).
- Leider, A. S., & Garbarino, V. E. (1987). Generalized hypercementosis. *Oral Surgery, Oral Medicine, Oral Pathology*, 63(3), 375-380. [https://doi.org/10.1016/0030-4220\(87\)90210-6](https://doi.org/10.1016/0030-4220(87)90210-6).
- Lustmann, J., & Bodner, L. (1988). Dentigerous cysts associated with supernumerary teeth. *International Journal of Oral and Maxillofacial Surgery*, 17(2), 100-102. [https://doi.org/10.1016/S0901-5027\(88\)80159-0](https://doi.org/10.1016/S0901-5027(88)80159-0).
- Manson-Hing, L. R. (2005). X-ray evidence of mechanical trauma. 1959. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology and Endodontics*, 100(2 suppl), S67-74. <https://doi.org/10.1016/j.tripleo.2005.05.001>.
- Marzook, H., Elhawary, Y., El-gendy, A., & Radwan, L. R. S. (2010). Cone Beam Computed Tomography Voxel Values Of Dental Structures. *Egyptian Dental Journal*, 56(4).
- Nunes, E., de Moraes, I. G., de Novaes, P. M. O., & de Sousa, S. M. G. (2002). Bilateral fusion of mandibular second molars with supernumerary teeth: case report. *Brazilian Dental Journal*, 13(2), 137-141. <https://doi.org/10.1590/s0103-64402002000200012>.
- Ochsenius, G., Escobar, E., Godoy, L., & Peñafiel, C. (2007). Odontogenic cysts: analysis of 2,944 cases in Chile. *Medicine Oral y Patologia Oral y Cirugia Bucal*, 12(2), E85-91.
- Santosh, P., Doni, B., Kaswan, S., & Rahman, F. (2013). Prevalence of dental anomalies in Indian population. *Journal of Clinical and Experimental Dentistry*, 5(4), e183-e186. <https://doi.org/10.4317/jced.51119>.
- Scarfe, W. C., Levin, M. D., Gane, D., & Farman, A. G. (2009). Use of cone beam computed tomography in endodontics. *International Journal of Dentistry*, 2009, 634567. Shoor, H., Sujir, N., Mutalik, S., & Pai, K. M. (2014). Hypercementosis: a rare finding in a patient with systemic lupus erythematosus. *BMJ case reports*, 2014, <https://doi.org/10.1136/bcr-2013-202370>.
- Sivakumar, T. T., Nair, B. J., & Raman, R. K. (2010). Double headed mandibular premolar – a miraculous developmental anomaly. *Oral & Maxillofacial Pathology Journal*, 1. <https://doi.org/10.1155/2009/634567>.
- Souza, L. N., Júnior, S. M. L., Pimenta, F. J. G. S., Souza, A. C. R. A., & Gomez, R. S. (2004). Atypical hypercementosis versus cementoblastoma. *Dento maxillo facial Radiology*, 33(4), 267-270. <https://doi.org/10.1259/dmfr/30077628>.
- Tunis, T. S., Sarne, O., Hershkovitz, I., Finkelstein, T., Pavlidi, A. M., Shapira, Y., Davidovitch, M., & Shpack, N. (2021). Dental Anomalies' Characteristics. *Diagnostics (Basel, Switzerland)*, 11(7), 1161. <https://doi.org/10.3390/diagnostics11071161>.
- Tabassum, S., & Khan, F. R. (2016). Failure of endodontic treatment: The usual suspects. *European Journal of Dentistry*, 10(1), 144-147. <https://doi.org/10.4103/1305-7456.175682>.



© 2024 by the authors. Submitted for possible open access publication under the terms and conditions of the Creative Commons Attribution (CC BY) license (<http://creativecommons.org/licenses/by/4.0/>).