Case Report

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Calcifying Hyperplastic Dental Follicle: A Rare Case Report

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Submitted 29 September 2025 **Accepted** 25 June 2026 **Published** 15 July 2026

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Citation

"Rai A, Keshwar S, Jain N, Raout T, Khadka S. Calcifying Hyperplastic Dental Follicle: A Rare Case Report. JPBKIHS. 2025;8(2):43-46."

https://doi.org/10.3126/jbpkihs.v8i2.84940

Abstract

Calcifying hyperplastic dental follicle is a rare condition characterized by impacted tooth associated with a hyperplastic dental follicle that include calcifications and odontogenic rests. Its features overlap with odontogenic cysts and tumors. Due to its rarity and overlapping characteristics, proper recognition of calcifying hyperplastic dental follicle is essential to avoid misdiagnosis. We report a case of a 22-year-old female who presented with an asymptomatic unerupted right maxillary canine detected during radiographic examination. Intraoral examination revealed absence of the right maxillary canine with normal overlying mucosa. The excised specimen was submitted for histopathological examination, which showed proliferating odontogenic epithelium arranged in islands, and areas of calcifications. These histopathological findings were consistent with a diagnosis of calcifying hyperplastic dental follicle.

Keywords: Calcification; Dental follicle; Impacted tooth

Declarations

Ethics approval and consent to participate: Informed consent was obtained from the patient.

Consent for publication: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Availability of data and materials: All data and materials supporting the conclusions of this case report are included within the published article.

Competing interest: None.

Funding Statement: None.

Authors' contributions: AR contributed to the manuscript preparation, literature review and data collection. SK (department in charge) contributed for the histopathological diagnosis and manuscript review. All authors read and reviewed the manuscript .

Acknowledgements: We would like to thank Department of Oral and Maxillofacial Surgery for their assistance during the management of this case.

INTRODUCTION

Calcifying hyperplastic dental follicle (CHDF) has been referred to as a hamartomatous growth of the dental follicle [1]. CHDF is found most frequently in third molars in the mandible and is mostly seen in young adults and adolescents [2]. However, involvement of other teeth, particularly maxillary anterior teeth, is uncommon and may represent an unusual anatomical presentation. Clinically, these lesions are usually asymptomatic and incidentally detected on radiographic examination for impacted teeth. Radiographically, they show as a well-circumscribed pericoronal radiolucency, mimicking a dentigerous cyst or other odontogenic lesion [2, 3].

Histologically, CHDF is characterized by fibrocellular stroma with odontogenic epithelial rests, multinucleated giant cells, and calcification foci including conglomerates of enameloid, cementoid, psammomatous or dystrophic calcifications [2, 3]. If multiple impacted teeth are affected with abundant calcifications and odontogenic epithelial rests, the condition has been termed multiple calcifying hyperplastic dental follicles (MCHDF), which may represent a distinct clinicopathological entity [4].

CASE DESCRIPTION

A 22-year-old female was referred to BPKIHS following the radiographic finding of an impacted tooth in right upper quadrant. There was no history of pain or other symptoms. The patient had a history of trauma to the upper anterior region seven years ago. Radiographic examination revealed an impacted right maxillary canine (13), suggesting failure of eruption rather than previous loss of this tooth. Patient had restoration of 22 and 23 two months back. The patient had no associated medical disorders.

On intraoral examination, the overlying oral mucosa clinically appeared normal, and the teeth 15, 14, 13, 12, 11 and 21 were absent. The orthopantomogram showed an unerupted mesially tilted right maxillary canine present on right upper quadrant with a well-defined pericoronal radiolucency of size 2-3 mm **(Fig. 1).** A provisional clinical diagnosis of dentigerous cyst was made.

An excisional biopsy was performed under local anesthesia. Tissue specimen was fixed in 10% formalin and sent for routine histopathological examination. On gross examination, single soft tissue specimen, white to light brown in color, with maximum dimension 10 x 8 x 2 mm3 in size was received. Microscopic examination showed odontogenic epithelium rests and areas of basophilic calcifications within the background of myxofibrous connective tissue and areas with high vascularity **(Fig. 2 and Fig. 3).** The calcified area consisted of acellular, small, basophilic and spherical bodies arranged in a whorled structure **(Fig. 4).** Tuft edges surrounding a few calcified structures were also observed **(Fig. 5).** Based on these histological findings, a diagnosis of calcifying hyperplastic dental follicle was established. Postoperative follow-up could not be documented as the patient was unable to report for further clinical evaluation.

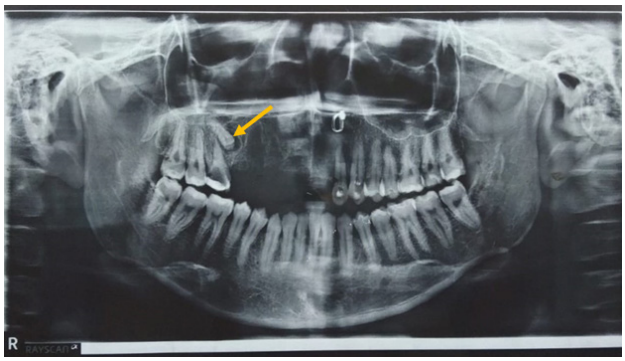

Figure 1: OPG shows peri-coronal radiolucency associated with medially impacted 13(arrow), and missing 15, 14, 12, 11 and 21.

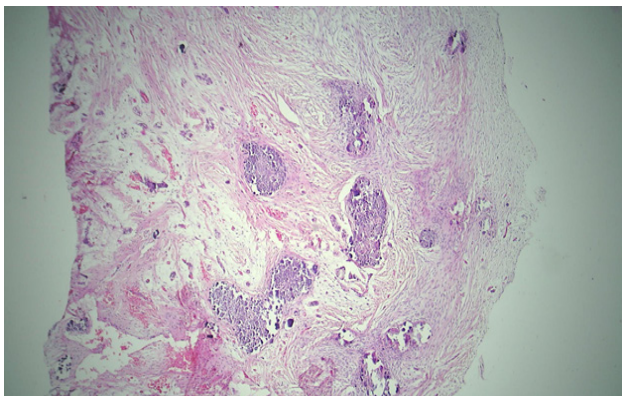

Figure 2:H& E; 4X areas of calcifications in fibro-cellular connective tissue with increase vascularity.

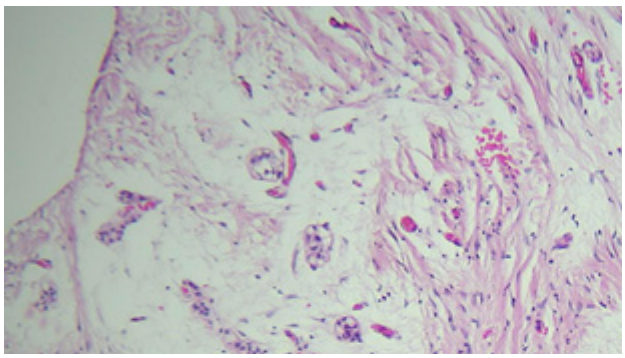

Figure 3: H&E; 10X shows odontogenic epithelial rests in fibro-myxoid connective tissue background.

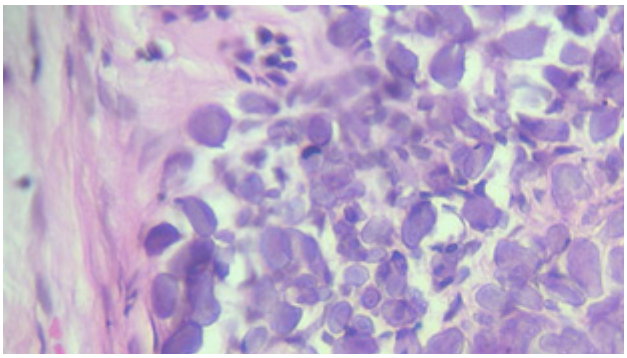

Figure 4: H&E; 40X type I calcifications having acellular, small, basophilic and spherical bodies arranged in whorled structure

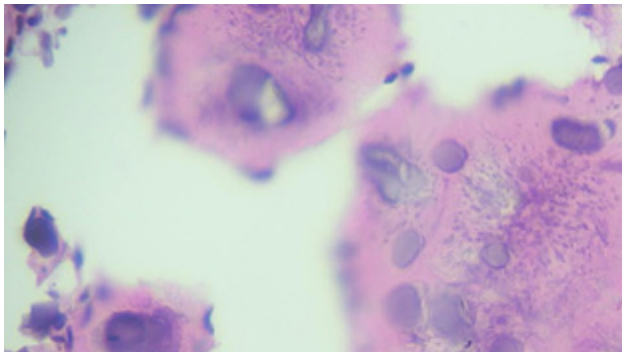

Figure 5: H&E; 40X Type II Tuft edges around few calcified structures.

DISCUSSION

The calcifying hyperplastic dental follicle is an odontogenic hamartomatous lesion arising in the pericoronal tissues of unerupted or impacted teeth. In the study of Hasegawa et al., 9 cases were reported, making CHDF a rare condition [2]. Previous reports have described CHDF commonly involving posterior teeth, especially mandibular third molars [2, 3]. The occurrence of CHDF around an unerupted upper canine, as observed in the present case, represents an uncommon presentation compared with the typical location described in the literature. The patient's history of trauma seven years prior, which resulted in the premature loss of adjacent teeth, provides a unique clinical context for considering whether long-standing post-traumatic tooth impaction may contribute to the development of follicular hamartomatous progression.

Clinically, it often affects young individuals, particularly in their second decade of life. Most reported cases are discovered incidentally during radiographic examination of impacted teeth [2, 3]. Microscopic findings in CHDF include the presence of fibrous connective tissue, odontogenic rests and areas of calcification [3]. Two types of calcifications have been described: type I and type II [5]. Type I calcification pattern shows acellular, small, basophilic and spherical bodies arranged in whorled structure. Type II calcification has spherular calcification pattern with diffuse peripheral fibrillar tufts [4, 5]. Type II calcification is fibrillar in nature, as demonstrated by polarized light [4]. In the present case, both types of calcifications are seen with a predominance of type I. The simultaneous coexistence of both Type I and Type II mineralization patterns within a solitary, non-multiple dental follicle is an unique histopathological finding that offers insights into the diverse calcifying potential of follicular mesenchymal cells.

In some cases, multiple impacted teeth and enlarged dental follicles that contain abundant calcifications and rests of odontogenic epithelium represent a distinct entity, termed as multiple calcifying hyperplastic dental follicles (MCHDF) [6]. Calcifying patterns of MCHDF are similar to those of CHDF [7]. A study done in 2024 by Hasegawa et al. described psammatoid calcification, calcifying whorled nodules (CWNs) and dentinoid deposits [2]. Immunohistochemically, the stromal spindle cells around CWN were positive for nestin and HES1 (Hairy and Enhancer of Split-1) [2]. The origin of calcifications could come from mesenchymal cells, which can differentiate into cementoblasts or odontoblasts under the influence of HES1 [2, 4].

Radiographically, CHDF mimics a dentigerous cyst, presenting as a pericoronal radiolucency. However, the radiolucency associated with dentigerous cyst is usually greater than 10 mm, and in CHDF there is no evidence of a cystic space between the soft tissue and the crown of the impacted teeth [4]. In our case, 2-3 mm of pericoronal radiolucency was seen. Histologically, dentigerous cyst shows a cystic cavity lined by 2-4 cell layers of non-keratinized stratified squamous epithelium [8].

Calcification patterns found in CHDF are also seen in the dental follicle of regional odonto-dysplasia [7]. Regional odontodysplasia can be distinguished from CHDF by radiographic examination, where regional odontodysplasia has an indistinct or fuzzy appearance of the coronal silhouette due lack of contrast seen between enamel and dentin, giving a 'ghost' appearance, and the presence of short roots [9]. Histopathologically, CHDF resembles calcifying epithelial odontogenic tumor (CEOT). CEOT is characterized by sheets and islands of polyhedral epithelial cells with prominent intercellular bridges, nuclear pleomorphism, and deposition of eosinophilic amyloid-like material (Congo red positivity with apple-green birefringence under polarized microscope) and concentric Liesegang ring calcifications. CEOT is an odontogenic tumor predominant in 4th decade characteristic "driven snow" radiopacities. In contrast, CHDF is typically

associated with younger patients, and there is absence of amyloid like material and Liesegang ring calcification. Immunohistochemically, CEOT often expresses odontogenic ameloblast associated protein and amyloid-related proteins, whereas CHDF lacks these markers [10]. By clearly delineating these explicit diagnostic boundaries, this report serves as a critical reference to prevent the misdiagnosis of CHDF as a more aggressive odontogenic neoplasm, ultimately sparing patients from unnecessary, over-aggressive surgical interventions.

CHDF is generally treated by removal of the associated tooth and enucleation of the follicular tissue. However, the lack of cystic fluid surrounding the pericoronal region is thought to render marsupialization inefficient. In some cases, eruption of the involved tooth has been achieved with orthodontic traction [7]. Prognosis is excellent, with no recurrence or malignant transformation reported [7].

CONCLUSION

The calcifying hyperplastic dental follicle is a rare odontogenic developmental anomaly that can closely mimic other odontogenic cysts and tumors both radiographically and histopathologically. The present case reports an atypical anatomical presentation involving an unerupted maxillary canine following long-standing post-traumatic impaction. Greater awareness and reporting of such cases will contribute to a better understanding of its clinical spectrum and distinction from other odontogenic pathologies.

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