Open Journal of
Clinical & Medical
Case Reports

Volume 8 (2022)
Issue 12

Case Report

ISSN: 2379-1039

Central odontogenic fibroma with a unique inverted funnel-shaped radiographic appearance

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Abstract

Odontogenic tumors are classified into three main groups: epithelial, mixed, and mesenchymal type based on interactions between odontogenic epithelium and ectomesenchyme. Odontogenic fibroma is a benign mesenchymal odontogenic neoplasm, which is an extremely uncommon neoplasm accounting only for 0.1% of all odontogenic tumors. Due to this tumor's variable presentation, and rarity, it is often confused with other tumors, and are usually not included in the routine differential diagnoses. Here, we report a case of central variant of odontogenic fibroma with an unique inverted funnel shaped radiographic appearance in an asymptomatic 14-year-old boy, with a chief concern of gaps between his teeth. Emphasizing the importance of including such lesions in the array of differential diagnoses when clinicians encounter such entities. The lesion was surgically enucleated completely with no evidence of recurrence. Histopathological examination confirmed the lesion to be a central variant of odontogenic fibroma. The boy is under orthodontic treatment for his concern.

Keywords

Odontogenic fibroma; Odontogenic tumor; Odontogenic mesenchymal tumor.

Introduction

Odontogenic tumors (OT) are classified into three main groups: epithelial, mixed, and mesenchymal type based on interactions between odontogenic epithelium and ectomesenchyme [1,2]. Odontogenic fibroma (OF) is a benign mesenchymal odontogenic neoplasm, evolving from the dental follicle, periodontal ligament, and dental papilla [3]. Lowell and colleagues in 1954, reported the earliest case of odontogenic fibroma, affecting a 38-year-old female [4]. Intraosseous (central) and peripheral variants have been recognized. It is an extremely uncommon benign neoplasm, accounting for approximately 0.1% of all odonto-

genic tumors [5,6]. Ana and colleagues in 2021, summarized the reported cases from different countries (Table 1) [7,8].

Table 1: Total number of reported central odontogenic fibroma cases [7].

Authors, year of publication	Country	Number of reported cases
Günhan, 1990	Turkey	18
Handlers, 1991	United States	19
Daley, 1994	Canada	25
Chong & Kok, 2000	Malaysia	46
Sriran & Shetty, 2008	India	12
Luo & Li, 2009	China	21
Eversole, 2011	United States	25
Mosqueda-Taylor, 2011	Mexico, Guatemala & Brazil	14
Zhou & Li, 2018	China	17

Central odontogenic fibroma (COF) presents as a slow-growing, asymptomatic swelling with expansion of cortical plates [5,9,10]. It occurs more commonly in the mandible, in the molar region, while the tumors of the maxilla are in the anterior region [2,5,9,11]. One-third of OFs are associated with unerupted teeth. Radiographically, smaller COF tumors present as a unilocular radiolucency with well-demarcated sclerotic borders, but larger tumors may appear multilocular [2,11]. The internal septa can vary from fine and straight to granular, resembling those seen in odontogenic myxomas or giant cell granulomas [9]. Histological examination is non-exclusive but usually reveals inactive odontogenic epithelial islands within collagenous stroma [12]. Due to this tumor's variable presentation and relative rarity, this lesion is often confused with other tumors, emphasizing the importance of correlation of clinical, radiographic, and histopathologic features. COF is reported to respond well to surgical enucleation, however, few cases have shown aggressive behaviour including recurrence [5,7,13–17].

Case Report

A 14-year-old male with a chief concern of space between his lower right front teeth reported to the Oral Health Centre, International Medical University, Kuala Lumpur, Malaysia. His parents noticed the gap at the age of 12 when the deciduous tooth exfoliated but the permanent tooth failed to erupt, although the tooth on the opposite side had erupted 2 years prior. There was no history of trauma, pain, discomfort, swelling, or discharge associated with the concern. His past medical, surgical, dental, family, and social histories were non-contributory. He desired to close up the gap for aesthetic purposes.

On extraoral examination, his face was symmetrical, without any noticeable swelling, and no palpable lymph nodes. Upon intraoral inspection, a full complement of permanent teeth except for all 3rd molars and the right mandibular canine were present. No obvious swelling or buccal obliteration was identified in the area of concern. The overlying mucosa was normal, and there was no tenderness on palpation (Figure 1). The teeth 41, 42, and 44 were positive for pulp sensibility tests and were firm, non-tender, with normal probing depths. Since the canine was missing, the patinet was subjected for further evaluations.

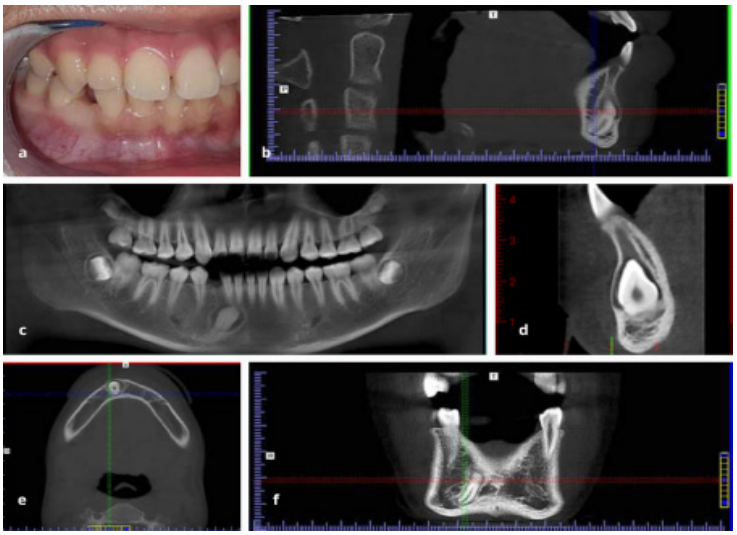

Figure 1: Figure 1a Intraoral photograph of the lateral right view showing a gap between #42 and #44; b-f Dental Panoramic radiograph, Axial, Coronal and Sagittal view of CBCT shows the position and angulation of impacted lower right mandibular canine with association of a well-defined unilocular radiolucency

A dental panoramic tomograph (DPT) (Figure 2) revealed an impacted tooth 43, apically below the roots of teeth 41 and 42. The crown and the root of the tooth 43 had completely formed. A unilocular pericoronal radiolucency with a well-demarcated corticated margin surrounding the crown of tooth 43 was noted. The radiolucency appeared to be attached to the tooth below the cementoenamel junction. Interestingly, the radiolucency was shaped like an inverted funnel, with a narrow opening extending coronally to the alveolar crest region between teeth 42 and 44.

A subsequent CBCT confirmed the findings observed on the DPT (Figure 3). A coronal view showed the crown of tooth 43 to be lingually tilted while the root towards the buccal surface and close to the lower border of the mandible. The sagittal view showed the inferior alveolar canal being displaced towards the lower border of the mandible. Considering the clinical and radiographic evidence, a differential diagnosis of a dentigerous cyst and odontogenic tumors like adenomatoid odontogenic tumor and ameloblastoma were considered.

An excisional biopsy was performed by anesthetizing the area of concern via local infiltration with 2% Xylonibsa (Lidocaine), with 1:100,000 epinephrine. A trapezoidal-shaped flap was raised to remove the overlying bone to expose the lesion. Intra-operatively, a soft tissue growth with a shape of an inverted funnel was noted. A complete enucleation together with its soft tissue pouch and surgical extraction of tooth 43 was performed. The alveolar mucosa at the superior margin of the lesion was excised to ensure complete removal of the lesion (Figure 2). Primary closure was performed using 4-0 Vicryl sutures. The specimen was immediately placed in 10% formalin and was sent for histopathological examination. Verbal and written post-operative instructions were provided. The patient was evaluated after one week, and the healing was satisfactory.

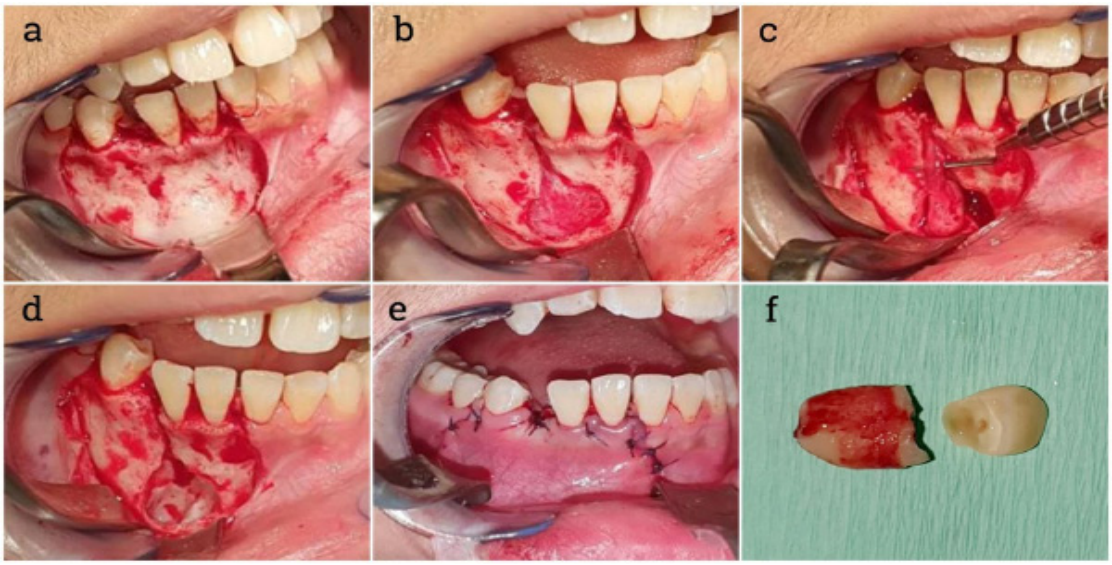

Figure 2: Intraoral photographs depicting the procedure of complete enucleation and surgical removal of #43. Note the inverted funnel shaped lesion.

On Histopathological examination, the microscopic sections stained with H and E showed a fibro myxoid connective-tissue with small scattered islands (nests and strands) of the odontogenic epithelium (Figure 3a,b). The islands were few to moderate in number and appeared inactive with no noticeable budding activity (Figure 3c,d). Odontogenic epithelial cells within a few islands also demonstrated clear vacuolated cytoplasm (Figure 3e,f). There were areas of dense engorged blood vessels (Figure 3h). Focal areas of connective tissue appeared myxoid, and the focal area of calcification within the stroma was also seen. The histopathological findings confirmed the lesion as a "Central Odontogenic fibroma." The histopathology report and the nature of the lesion were explained to reassure the patient. The patient was then referred back to the orthodontist for closure of the gap.

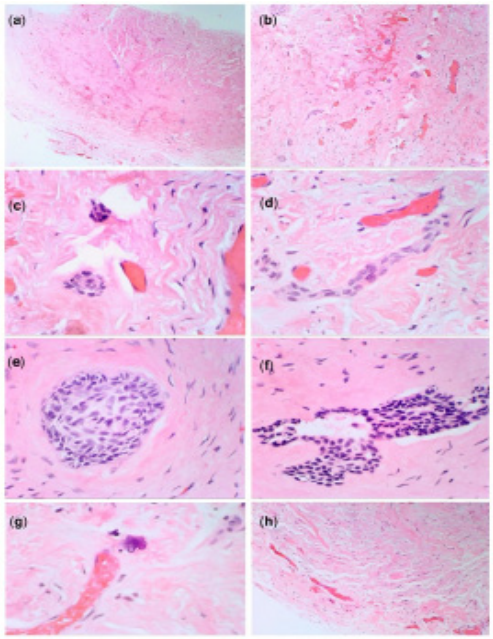

Figure 3: Histopathological examination reveals a fibro myxoid connective-tissue with small scattered islands (nests and strands) of the odontogenic epithelium (Figure 3a,b). The islands were few to moderate in number and appeared inactive with no noticeable budding activity(Figure 3c,d). Odontogenic epithelial cells within a few islands also demonstrated clear vacuolated cytoplasm (Figure 3e,f). There were areas of dense engorged blood vessels (Figure 3h).

Discussion

Although OF was first described in 1971 by WHO, the definition was not concordant, and this lasted till 2005. The WHO classification in 2005 defined OF as "a rare neoplasm characterized by varying amounts of inactive-looking odontogenic epithelium, embedded in a mature, fibrous stroma"[18]. It was subdivided into two groups based on histological criteria; the epithelial-poor type (simple type) and an epithelial-rich type (complex or WHO type) [2,19]. After extensive debates, this subclassification was abandoned as its definition and documentation were insufficient since there was no clinical significance between them [2,18,19]. The lesion is now categorized based on its location; central or peripheral [11,18]. Peripheral OF is the most common peripheral OT, accounting for approximately 51.1% to 63.6% of all cases [20,21]. On the contrary, COF is a rare tumor accounting for 1.5% of central OTs. To date, only 135 cases of COF have been reported in the literature [18].

The radiological diagnosis in our case was tough as the lesion was associated with an unerupted mandibular canine, resembling a dentigerous cyst. It was further complicated by the shape of the radiolucency, mimicking an inverted funnel-shape with a narrow opening. Although uncommon, similar cases of unilocular radiolucency associated with an impacted tooth are reported [11,22]. However, this unique radiolucency can also be attributed to the presence of gubernacular cord which is a normal anatomical structure that plays a role in tooth eruption by connecting the dental follicle of the permanent tooth to the connective tissue of the oral epithelium. More extensive evaluations of 3D radiographs involving impacted teeth may help shed light on this understanding.

A wide range of differential diagnoses such as COF, dentigerous cyst, odontogenic keratocyst, ameloblastoma, desmoplastic fibroma, and ameloblastic fibroma should be considered when associated with an impacted tooth. On contrary, when not associated with an impacted tooth, the list could be expanded to entities under the umbrella of odontogenic cysts and tumors, including odontogenic myxoma [18].

The COF responds favourably to surgical enucleation [7,28]. In more complicated cases, adjunctive treatment such as tooth extractions and even endodontic treatments [18] may be required. Conservative surgical approaches such as curettage are less commonly practiced [18,23]. Although uncommon, COF recurrences have been reported, with a recurrence rate of 6%, and are usually associated with the inadequate curettage of the lesion and not the histologic type [18,28]. There have been no reports of malignant transformation of COF but long-term follow-up is advised [7,12,18,28]. In the present case, there have not been any recurrence 20 months after surgery, and the patient is currently under orthodontic treatment.

Conclusion

We present a rare case of COF with an unusual radiographic appearance, with its shape resembling an inverted funnel and a unique association with an impacted mandibular canine tooth. COF is a rare benign odontogenic tumor, that has an inconsistent clinical and radiographic presentation, imitating other lesions and complicating the diagnostic algorithm. With this, we highlight the importance of including COF in the

differential diagnosis of intra-bony tumors affecting the jaws.

Declarations

Conflict of interest: The authors declare no conflict of interests.

Consent: Written consent is obtained from the patient for publication purpose.

References

1. Speight PM, Takata T. New tumour entities in the 4th edition of the World Health Organization Classification of Head and Neck tumours: odontogenic and maxillofacial bone tumours. Virchows Archiv. 2018; 472.

2. Wright JM, Soluk Tekkeşin M. ODONTOGENIC TUMORS. WHERE ARE WE IN 2017? Journal of Istanbul University Faculty of Dentistry. 2017; 51.

3. Zia M, Arshad A, Zaheer Z. Central Odontogenic Fibroma: A Case Report. Cureus. 2018.

4. Pincock LD, Bruce KW. Odontogenic fibroma. Oral Surgery, Oral Medicine, Oral Pathology. 1954; 7.

5. Ayesha RT, Pachipulusu B, Govindraju P, Ravindra S. Odontogenic Fibroma Associated with an Impacted Tooth. JOURNAL OF CLINICAL AND DIAGNOSTIC RESEARCH. Published online 2018.

6. Spencer John, Daniels M. Central odontogenic fibroma of mandible: a case report and review of the literature. al Med Oral Pathol Oral Radiol Endod. 2004; 3: 295-300.

7. Roza ALOC, Sousa EM, Leite AA, et al. Central odontogenic fibroma: an international multicentric study of 62 cases. Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology. 2021; 131.

8. Siar CH, Ng KH. Clinicopathological study of peripheral odontogenic fibromas (WHO-type) in Malaysians (1967–95). British Journal of Oral and Maxillofacial Surgery. 2000; 38.

9. Balaji P GAPGVVJU. Central Odontogenic Fibroma of Maxilla: A Rare Case. International Journal of Scientific Study. 2015; 2: 189-192.

10. Murgod S, Girish H, Savita J, Varsha V. Concurrent central odontogenic fibroma and dentigerous cyst in the maxilla: A rare case report. Journal of Oral and Maxillofacial Pathology. 2017; 21.

11. Santoro A, Pannone G, Ramaglia L, Bufo P, lo Muzio L, Saviano R. Central odontogenic fibroma of the mandible: A case report with diagnostic considerations. Annals of Medicine and Surgery. 2016; 5.

12. Harti K, Oujilal A, Wafaa EW. Central odontogenic fibroma of the maxilla. Indian Journal of Dentistry. 2015; 6.

13. Seo Y-K, Kang JH, Lee SR, Choi Y-S, Hwang E-H, Oh SH. Diagnostic considerations in central odontogenic fibroma of the maxilla: 2 case reports. Imaging Science in Dentistry. 2019; 49.

14. Kinney LA, Bradford J, Cohen M, Glickman RS. The aggressive odontogenic fibroma: Report of a case. Journal of Oral and Maxillofacial Surgery. 1993; 51.

15. Reichart PA, Philipsen HP, Moegelin A, Thalmann U. Central odontogenic fibroma, granular cell variant. Oral Oncology Extra. 2006; 42.

16. Sachdeva SureshK, Verma P, Verma D, Verma K. Recurrent central odontogenic fibroma: An uncommon clinical presentation. Saudi Surgical Journal. 2014; 2.

17. CARVALHO Stephany P., WATANABE Satiro, VENCIO. Eneida F. A RECURRENT CASE OF CENTRAL ODONTOGENIC FIBROMA IN MAXILLA SHOWING ROOT RESORPTION: A CASE REPORT. Caso Clínico. 2018; 27: 40-43.

18. Bennabi S, Lesclous P, Cloitre A. Central Odontogenic Fibroma: characteristics and management. Journal of Oral Medicine and Oral Surgery. 2021; 27.

19. Wright JM, Vered M. Update from the 4th Edition of the World Health Organization Classification of Head and Neck Tumours: Odontogenic and Maxillofacial Bone Tumors. Head and Neck Pathology. 2017; 11.

20. Luana Samara B. de-Sena, Márcia Cristina C. Miguel, Jozinete V. Pereira, Daliana Q. C. Gomes, Pollianna M. Alves, Cassiano Francisco W. Nonaka. Peripheral odontogenic fibroma in the mandibular gingiva: case report. J Bras Patol Med Lab. 2019; 55: 192-201.

21. Baiju C, Rohatgi S. Peripheral odontogenic fibroma: A case report and review. Journal of Indian Society of Periodontology. 2011; 15.

22. Pippi R, Santoro M, Patini R. The central odontogenic fibroma: How difficult can be making a preliminary diagnosis. Journal of Clinical and Experimental Dentistry. 2016.

23. Pushpanshu Kumar, Kaushik rachana, Punyani R Silky, Jsuaja Vishal, Raj Vineet, Seshadri Abitha. Concurrent central odontogenic fibroma (WHO Type) and traumatic bone cyst: report of a rare case. Quantitative Imaging in Medicine and Surgery. 2013; 3: 341-346.

Manuscript Information: Received: July 16, 2022; Accepted: August 17, 2022; Published: August 19, 2022

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Citation: Ravi Banavar S, Nga JDL, Foo SYT, Khoo SP, Jnanendrappa N. Central odontogenic fibroma with a unique inverted funnel-shaped radiographic appearance. Open J Clin Med Case Rep. 2022; 1892.