ISSN: 0976-3031

Available Online at http://www.recentscientific.com

CODEN: IJRSFP (USA)

International Journal of Recent Scientific Research
Vol. 9, Issue, 2(H), pp. 24287-24290, February, 2018

International Journal of Recent Scientific Research

DOI: 10.24327/IJRSR

Case Report

AN UNUSUAL CASE OF AN INFECTED DENTIGEROUS CYST-A CLINICAL CONUNDRUM

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DOI: http://dx.doi.org/10.24327/ijrsr.2018.0902.1634

ARTICLE INFO

Article History:
Received 16th November, 2017
Received in revised form 25th December, 2017
Accepted 23rd January, 2018
Published online 28th February, 2018

Key Words:
Dentigerous Cyst, Unerupted Tooth, Mandibular Osteotomy.

ABSTRACT

A dentigerous cyst or follicular cyst is a form of odontogenic cyst. The cyst is associated with the crown of the impacted or unerupted teeth. It usually remains completely asymptomatic unless when infected and can be discovered only on routine radiographic examination. However, when infected it may turn painful, grow in size and result in a palpable mass. Additionally, as they grow they displace adjacent teeth and result in severe bone destruction. Although, hyperplastic follicular tissue and dentigerous cyst are the two most common entities associated with the crown of an impacted tooth, other diagnostic possibilities, including odontogenic tumors should also be considered in case of a pericoronal radiolucency. This article presents a case of an infected dentigerous cyst which mimicked other cysts and tumors and was quite a challenge in terms of an accurate diagnosis, yet despite timely interventions had horrendous consequences in terms of prognosis.

INTRODUCTION

Dentigerous cyst (DC) is one of the most common lesions of the jaw and one of the most common type of odontogenic cyst [1]. DC is one that encloses the crown of an unerupted tooth by expansion of the follicle and is attached to its neck. The term dentigerous is preferred, the literal meaning being 'tooth bearing.' In order of its frequency, they are associated mandibular third molars, maxillary canines, mandibular second premolars and maxillary third molars [2]. Hereby, we present a case of a 45-year-old female patient who had reported to our department with an extra-oral swelling with just one month's history.

Case Report

A 45-year old female patient reported to our department with chronic pain and associated swelling on the right side of the face since a duration of one month and 15 days respectively. Her medical and family histories were non-contributory. Patient gave a history of previous extraction of teeth in lower jaw. On extra-oral examination there was a solitary, hard swelling with diffuse borders extending from above the ala-tragus line to below the lower border of the mandible on the right side [Figure 1-A]. On intra-oral examination the mouth opening was restricted with blanching of the oral mucosa, 35 and 45 were missing with root stump in relation to 46 [Figure 1-B]. There was obliteration of the buccal vestibule on the right side and was tender to touch. A provisional diagnosis of buccal space infection due to periapical abscess in relation to 46 was established and as intra-oral radiograph was not possible ailing to the lack of adequate mouth-opening, a mandibular occlusal radiograph followed by an orthopantomogram (OPG) was carried out. An incision was given and pus was drained followed by placement of sutures [Figure 1-C].

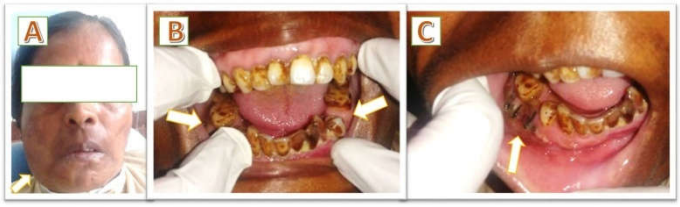

Figure 1 A-Unilateral Extra-Oral Swelling Causing Gross Facial Asymmetry. **B**-Blanching of Oral Mucosa. **C**-Sutures Placed After Drainage of Pus Intra-Orally.

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A multi-locular radiolucency with radiopaque flecks were observed on the radiographic images [Figure 2 - A] extending from region of 35, crossing the midline and extending beyond region of 48 involving two-third of the ramus of the mandible on the right side, also a pathological fracture was observed along the lower border of mandible in the region below 48 [Figure 2-B]. An incisional biopsy was undertaken following which the patient was advised for a cone beam computed tomography (CBCT) to get finer details of the extent of the lesion. CBCT images revealed the pathological fracture and extent of the lesion measuring about over 100 mm in length across the mandible and about 21 mm in height [Figure 3-A, B]. While the bucco-lingual extent of the lesion on axial section was about 19 mm and the cross-section images showed the extent of radiolucency to be about 15 mm in diameter, also the exact orientation of the impacted pre-molar could also be well appreciated [Figure 3-C, D].

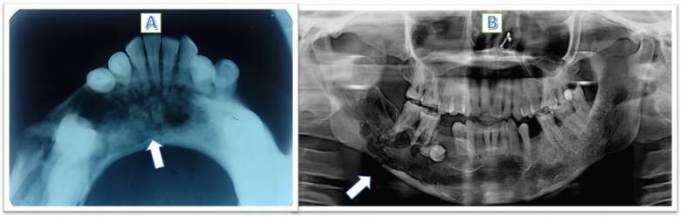

Figure 2 A-Mandibular Occlusal Radiograph Showing Multilocular Radiolucency and Impacted Pre-Molar. **B**-OPG Showing the Extent of the Radiolucent Lesion.

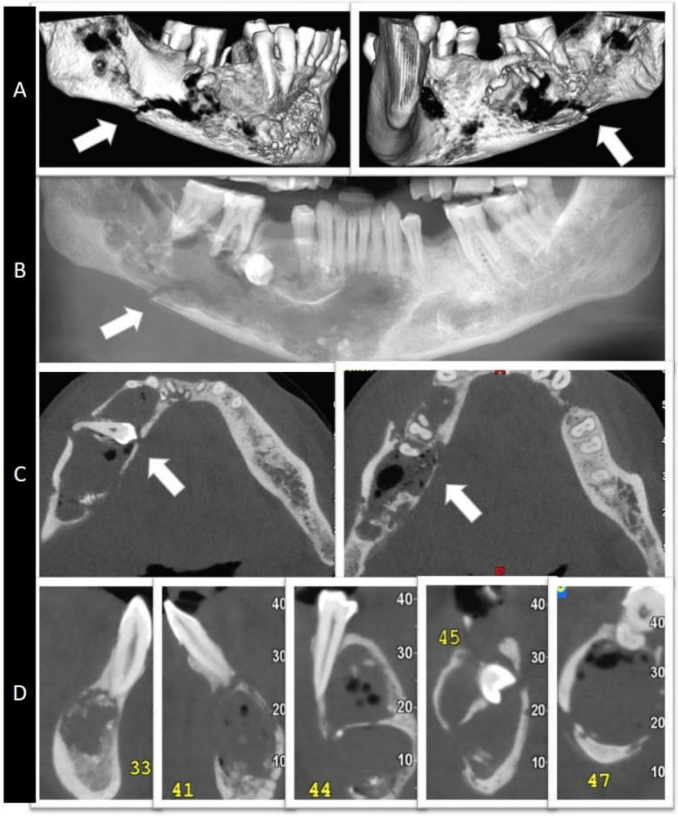

Figure 3 The Extent of the Lesion Well-Appreciated On The CBCT Images; **A**-In 3-Dimesional Images, **B**-In Panoramic Image, **C**-On Axial Sections, **D**-On Cross-Sectional Images of Teeth 33, 41, 44, 45 And 47.

Based on all the radiographic findings: dentigerous cyst, ameloblastoma and keratocystic odontogenic tumor were prinicipally considered as the radiographic differential diagnosis. The initial biopsy report showed connective tissue with inflammatory cell infiltration suggestive of a non-specific inflammatory tissue [Figure 4-A, B]. So a second biopsy was undertaken which revealed fibrous connective tissue, chronic inflammatory cells and reduced enamel epithelium confirming the final diagnosis as an infected dentigerous cyst. Hemimandibulectomy of the right side was carried out with free fibula flap reconstruction. But, post-surgery the recovery phase was eventful and lead to various medical complications which resulted in an untimely demise of the patient.

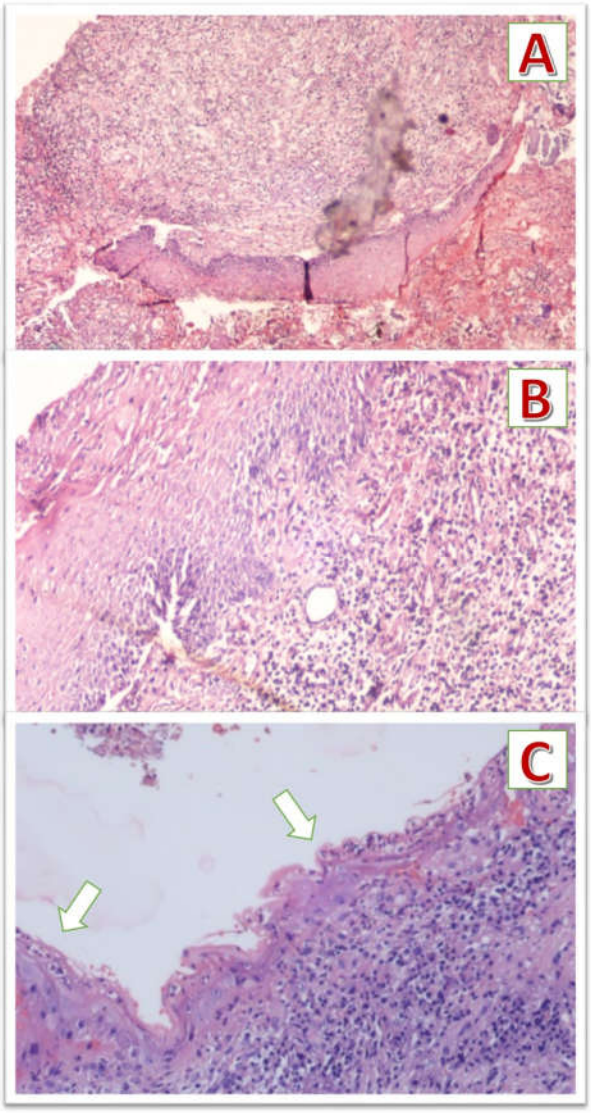

Figure 4 Histological Photomicrograph Showing - Non-Specific Inflammatory Tissues: **A -** (H & E - 4x). **B**-(H & E - 10x). **C**-Infected Dentigerous Cyst (H & E - 10x).

DISCUSSION

A DC is an odontogenic cyst associated with the crown of an unerupted or partially erupted tooth. The cyst cavity is lined by epithelial cells derived from the reduced enamel epithelium of the tooth forming organ [2]. DCs were earlier termed as 'Follicular cysts' since it was assumed that these cysts were derived from tooth follicle which is a mesodermal structure. Later this term was abandoned as it was conceived on an erroneous perception. Literature reveals that DC constitute more than a quarter of all jaw cysts with a very slight male preponderance. [3]

Predominating occurrence is the second to third decade of life. About 70% of dentigerous cysts occur in the mandible and 30% in the maxilla [4]. Most DC are solitary. Multiple/bilateral cysts are usually found in association with a number of syndromes including cleidocranial dysplasia, Maroteaux--Lamy syndrome and Gorlin--Goltz syndrome [5].

Usually all DCs arise from the enamel organ after completion of amelogenesis. Some of the prominent theories of DC formation are listed below: [3,6]

- Intrafollicular theory: The formation occurs due to fluid accumulation between the layers of inner and outer enamel epithelium after crown formation.
- Enamel hypoplasia theory: The formation occurs due to degeneration of stellate reticulum at a very early stage of tooth development.
- Main's theory: It suggests that impacted tooth exerts pressure on the follicle with resulting obstruction of venous outflow. This induces rapid transudation of fluid across the capillary walls.

DCs are usually small asymptomatic lesions that are an incidental finding on routine radiographs; hence, when the cyst is smaller in size, it would be difficult to differentiate it from a larger but normal dental follicle. A working definition to rule out this radiographic confusion is that, a DC exists only when the distance between the crown and dental follicle is greater than 2.5 to 3.0 mm [7]. Patients usually present with painless slow growing swelling involving the affected area. This swelling is very firm on palpation indicating cortical expansion as seen in our case.

The differential diagnosis must include lesions such as keratocyst, primordial cyst and odontogenic tumors such as mural ameloblastoma, unilocular ameloblastoma, ameloblastic fibroma and adenomatoid odontogenic tumor. Bilateral DCs should raise suspicion of a syndrome or systemic condition [8].
A periapical inflammation of the non-vital deciduous teeth or adjacent permanent teeth has been suggested as a factor triggering the formation of an inflammatory/infected DC (IDC) of the unerupted permanent successors. Benn and Altini considered three possible mechanisms in the histogenesis of IDCs: [7]

- Intrafollicular DCs formed around the crowns of permanent tooth that become secondarily inflamed, as a result of periapical inflammation spreading from nonvital deciduous predecessors.
- Radicular cysts at apices of non-vital deciduous teeth or adjacent permanent teeth that fuse with the follicles of unerupted permanent successors.
- Periapical inflammation from any source, but usually from non-vital deciduous teeth spreading to involve follicles of unerupted permanent successors.

Therefore, the appearance of IDC is most commonly found across literature involving the mixed dentition stage as the spread from non-vital deciduous teeth is more likely making our case distinctly rare.

In plain radiographs these cysts present as a well-defined unilocular radiolucency. Often there is a demarcating sclerotic border. A large DC may provide an impression as if it is multilocular. This appearance is due to the persistence of bony trabeculae within the radiolucency as seen in our case. But, these cysts are particularly unilocular in nature [3].

Three types of DCs have been described radiographically: The central variety, in which the radiolucency surrounds just the crown of the tooth, with the crown projecting into the cyst lumen. In the lateral variety, the cyst develops laterally along the tooth root and partially surrounds the crown and the circumferential variant exists when the cyst surrounds the crown but also extends down along the root surface as if the entire tooth is located within the cyst [2]. Our case radiographically, was most likely a classic presentation of exacerbation of the circumferential variety.

Methods employed for elimination of DC have included decompression, marsupialisation, and enucleation. However, the criteria for selecting these treatment modalities (indications and contraindications) are not clearly defined. Surgery is usually recommended for a DC, and consists of enucleation and extraction of the teeth embedded in it or affected by it. In very large cysts, an initial phase of marsupialization of the lesion to the oral cavity followed by the usual surgical treatment is also recommended [9].

Possible complications of this cyst include: permanent bone deformation, resorption of mandibular canal wall, displacement of secondary teeth, or development of an ameloblastoma or epidermoid carcinoma. Hence early diagnosis and removal of a DC is imperative to reduce morbidity and at times even mortality as in our case [10].

CONCLUSION

DCs are mostly asymptomatic. But if turned infected in such cases when left untreated, it can spread extensively in minimal time and result in grave consequences. Higher imaging modalities and knowledge of histopathology can indeed help us in effective diagnosis for when infected it can mimic various other pathologies. Thus, "Ignorance ain't blissful, and knowledge is truly wealth." as it helps in early diagnosis and timely interventions, for they are of utmost importance to dictate a good prognosis.

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How to cite this article:

Geon Pauly *et al*.2018, An Unusual Case of An Infected Dentigerous Cyst-A Clinical Conundrum. *Int J Recent Sci Res.* 9(2), pp. 24287-24290. DOI: http://dx.doi.org/10.24327/ijrsr.2018.0902.1634

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