ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)

IJDSIR : Dental Publication Service

Available Online at: www.ijdsir.com

Volume – 2, Issue – 2, March - April - 2019, Page No. : 78 - 83

Biopsy of an Innocent Swelling Resulting in a Rare and Unusual Clinical Diagnosis

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Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Intraoral lesions often present as nodules or swellings. A soft tissue swelling can involve any region of the oral mucosa including gingiva. The primary disease processes that give rise to swellings and tumors of the oral cavity include cysts, mucous extravasation and retention in the minor salivary glands, foci of granulation tissue and inflammation, abscesses, and connective-tissue proliferations that are well-defined or encapsulated, as well as infiltrative sarcomas. Odontogenic swellings are of great concern as they affect the oral cavity involving the bone or soft tissue. Many intraoral swellings may have similar clinical presentation; therefore microscopic examination is essential inorder to arrive at a final diagnosis. This article discusses a case where a non-suspicious gingival swelling in a teenaged male patient turned out to be peripheral unicystic ameloblastoma.

Keywords: Gingiva, Biopsy, Ameloblastoma, Neoplasm.

Introduction

Oral soft tissues are affected by a multitude of pathologic conditions of variable etiology and significance and their appropriate management relies on their accurate diagnosis. Overlapping of the signs and symptoms pose significant problems for their diagnosis, which can be resolved only through a thorough knowledge of the clinicopathologic characteristics of each condition and a systematic approach to diagnosis. An essential component of the diagnostic process is the formulation of a differential diagnosis. The goal of differential diagnosis is to determine the nature of the enlargement as a basis for formulating a rational treatment approach.[1] The

symptoms, growth rate, palpation characteristics, surface morphology, and lesion site allow for categorization of the soft tissue lesions into one of the five lesion groups that includes: papillary surface enlargements, acute inflammatory enlargements, reactive hyperplasia, benign submucosal cysts and neoplasms, and aggressive and malignant neoplasms. Bony enlargements of the maxilla and mandible are divided into three categories: inflammatory lesions, benign cystic and neoplastic lesions, and aggressive and malignant lesions.[2]

One of the best known epithelial odontogenic tumours is Ameloblastoma. It is benign in nature, locally aggressive with an insidious growth pattern and exhibit diverse clinical, radiographic and histological patterns. They may originate from the rests of dental lamina, enamel organ, lining or walls of non-neoplastic odontogenic cyst and nevertheless also from the basal layer of oral epithelium.[3]

Unicystic ameloblastoma is a term that is derived from its macroscopic and microscopic appearance of being presenting as a large monocystic cavity with a lining that is focally ameloblastomatous. The concept of Unicystic Ameloblastoma was first introduced by Robinson and Martinez in 1977.[4] Despite the term "unicystic", radiographically, the lesion not only appears unilocular but also as a multilocular defect in the jaw bones.[5]

Peripheral Ameloblastoma (PA) also known as extraosseous ameloblastoma, is a rare variant and exhibits the same histological characteristics as solid/multicystic ameloblastoma. Kuru in 1911 first reported PA, but it was Stanely and Krogh in 1959 who first defined the clinical and histopathological characteristics of this lesion. As to the location of PA, the maxilla/mandible ratio is 1:2.6. The mandibular premolar region accounts for 32.6% of all sites.[6]

This article discusses a rare case of peripheral Unicystic Ameloblastoma in a 17-year old male patient. This is a case of clinical interest because of its rare and asymptomatic occurrence in the gingiva overlying unerupted right mandibular third molar in a young individual and higher recurrence rate.

Case Report

A 17-year-old male patient reported to the department of Periodontics with a chief complaint of gum overgrowth in lower right back tooth region since one year. On examination, patient was systemically healthy. There was no history of trauma or infection. He gave history of initially small gingival overgrowth that gradually increased to present size. Not associated with pain or extraoral swelling. On intraoral examination, the growth was seen in the mandibular right posterior tooth region covering the unerupted third molar and half of the occlusal surface of second molar. It was brownish pink in colour, 10mm long and 5mm wide (Figure 1). On palpation, it was non-tender, sessile with a smooth surface, firm in consistency with well defined margins. Based on the patient's chief complaint and clinical examination, the swelling was provisionally diagnosed as an irritation fibroma. Examination of orthopantomograph revealed an unerupted third molar with blunderbuss canals (Figure 2).

The growth was carefully excised under local anesthesia using No. 15 surgical blade. Proper aseptic measures were taken (Figure 3). After excision, the tissue was thoroughly examined and sent for histopathological examination (Figure 4).

Histopathological Report

Hematoxylin an Eosin section showed presence of odontogenic cystic epithelial lining within the connective tissue stroma. The lining epithelium showed the presence of tall columnar palisading basal cells (Figure 5) with hyperchromatic nuclei resembling ameloblast like cells (Figure 6) and loosely arranged stellate reticulum like cells in supra basal layers (Figure 7a, 7b). Stellate

reticulum like cells were of variable thickness exhibiting thickened and highly proliferating areas. The epithelial lining also showed the presence of squamous metaplasia in few areas and showed vacuolated cells in the superficial layer. There was presence of both active and inactive odontogenic epithelial islands within the connective tissue stroma. Parakeratinised stratified squamous surface epithelium of variable thickness was present. The connective tissue stroma showed the presence of loose collagen fiber bundles, fibroblasts, endothelial-lined blood vessels with endothelial cell proliferation and mild inflammatory cell infiltrate chiefly composed of lymphocytes.

A final diagnosis of Peripheral Unicystic Ameloblastoma was given. At 10th day examination healing of surgical site was satisfactory (Figure 8). The patient was subsequently recalled after 6 months and one year (Figure 9). CT scan of the region, performed 1 year after excision of the lesion, did not reveal any involvement of underlying bone.

Discussion

Peripheral Ameloblastoma is a rare lesion and accounts for 2 to 10 percent of all ameloblastomas.[7] The lesion resembles epulis and other benign tumours like pyogenic granuloma, peripheral gaint cell granuloma more commonly. It is sessile with exophytic growth restricted to soft tissues overlying tooth bearing areas of jaws. The male to female predilection ratio is 1.9:1 with more predilections for the male. Zhu *et al.*[8] in a review of 43 Japanese cases, reported that PA is more common in 5th and 7th decades with 70% of lesions occurred in males and most common site is mandibular premolar region. El-Hakim and El-khashab reported an unusual case of PA in conjunction with UA in a 13-year-old male child, where the lesion appeared to develop from lining of dentigerous cyst related to an impacted mandibular canine.[9]

The histogenesis of PA is controversial. Two main theories have been developed about the cellular origin of PA. Some tumors are completely located within the gingival connective tissue with no contact with the surface epithelium or some demarcated from surface epithelium by a connective tissue band which are called glands of serres. Other theory stated that PA is very close to surface epithelium.[6]

Bone involvement in PA is absent with the presence of small depression on the bone surface which is known as cupping or saucerisation.[10] The lack of infiltration can be explained by the presence of fibrous barrier surrounding the lesion from gingiva and periosteum. This behaviour of PA makes it different from the intraosseous ameloblastoma where a high degree of bony erosion and marrow infiltration can be seen.

The treatment of this type of lesion remains controversial and it is based on recurrence and aggressiveness. The choice of treatment depends on the microscopic pattern, location and size of the lesion, age of the patient. The treatment of choice is conservative local excision without removing bone or teeth. In the present case, the growth appears to be sessile. Intraoral radiograph, orthopantomogram and computed tomography scan did not show any bone destruction. As the recurrences of PA ranges from 16% to 19% long term follow up was planned.[11]

This case is considered to be rare and is of clinical interest because it has occurred in a very young 17-year-old male patient, literature suggests 5th and 7th decades to be common ages of occurrence and it has occurred in a relatively uncommon region such as mandibular third molar, literature suggests mandibular premolar region as the common area of occurrence[6].

Conclusion

PA is an uncommon odontogenic neoplasm. A careful histopathological evaluation is necessary to differentiate it from other lesions of oral cavity, and all possible cases should be evaluated by immunohistochemistry. Although conservative treatment is preferred, long-term follow-up of patients is mandatory to prevent recurrence of lesion and for better prognosis. The lesion described in this article was evaluated clinically, excised conservatively, diagnosed histopathologically and followed up over a period of one year.

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Legends

Fig 1: Gingival swelling overlying unerupted 48 & covering occlusal surface of 47

Fig 2: Preoperative Orthopantomograph

Fig 3: Immediate postoperative

Fig 4: Excised tissue (10×5mm)

Fig 5: Tall columnar cells

Fig 6: Ameloblastic cells extending into cystic space

Fig 7a: Stellate reticulum cells under low power magnification

Fig 7b: Stellate reticulum cells under high power magnification

Fig 8: 10th day Post operative

Fig 9: One year Post operative

Figures

Fig1:

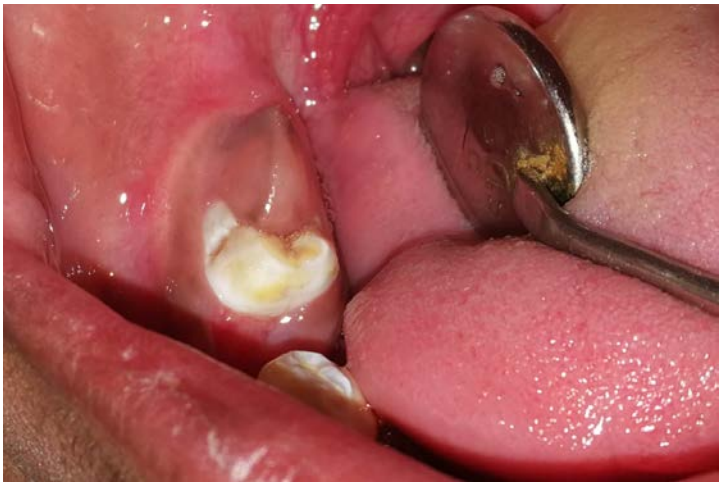


Fig2

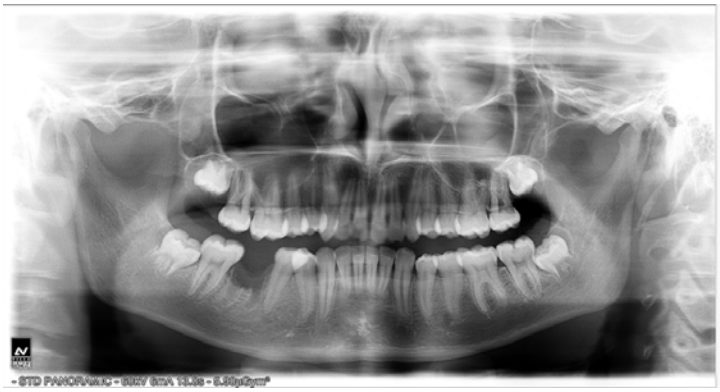
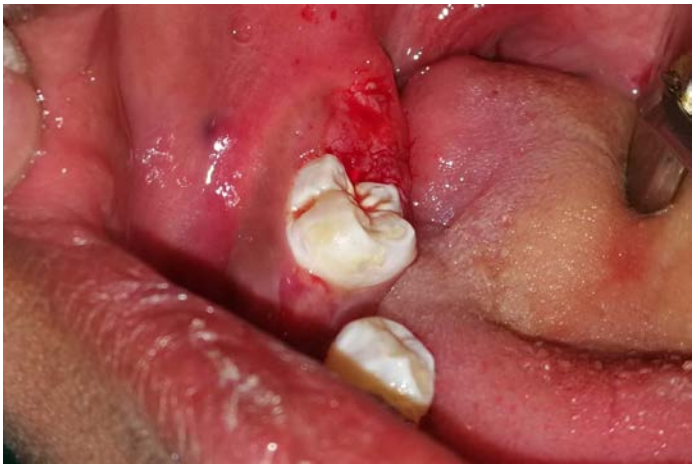


Fig3:



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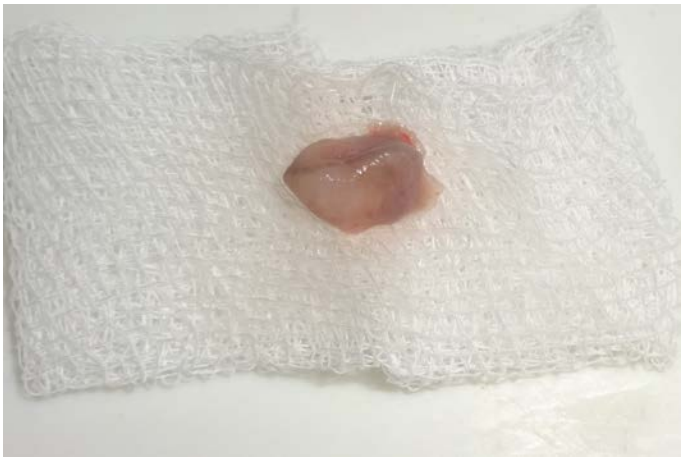

Fig5

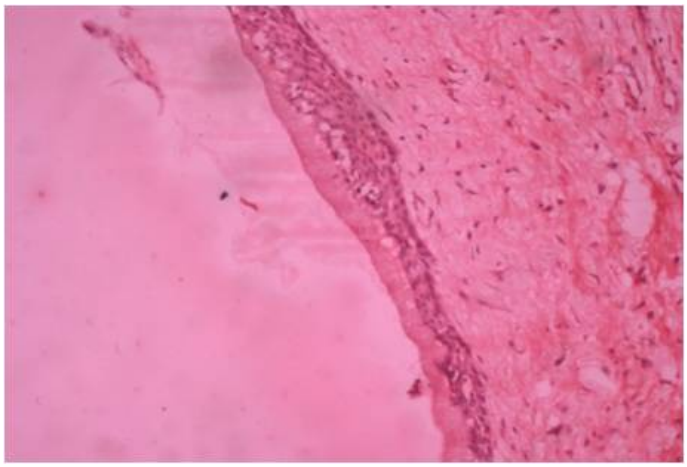

Fig6

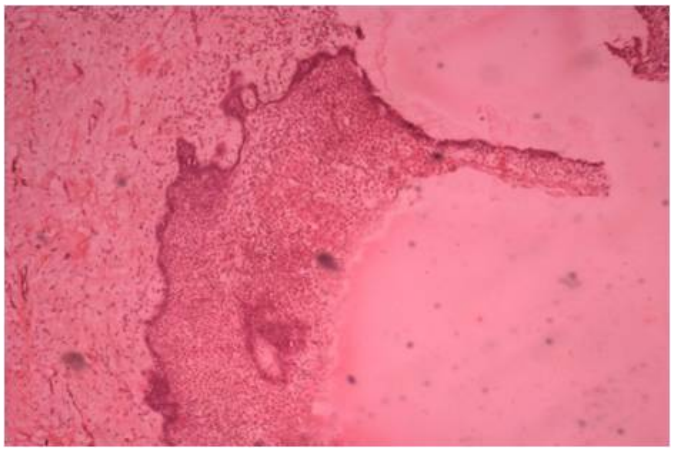

Fig7a

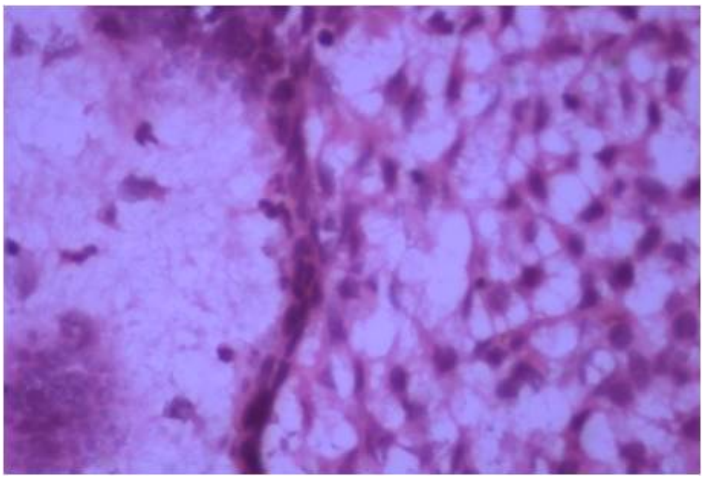

Fig7b

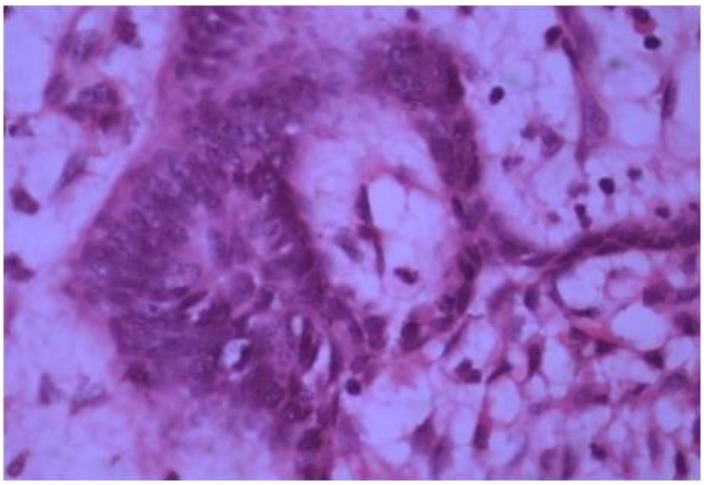

Fig8

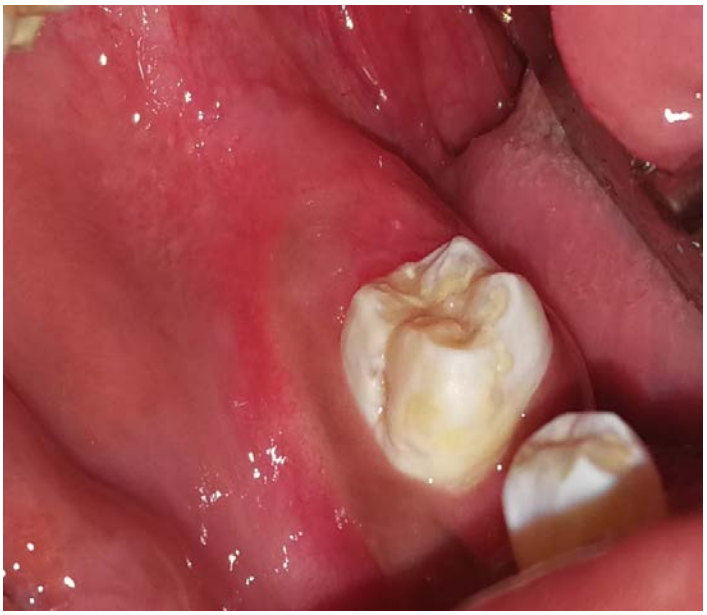

Fig9