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The Occurrence of Jaw Lesions Associated with Impacted Teeth Could be familial? A Case Report

Dr. Saranya Varadarajan[1*], Dr. Sujatha Govindarajan[2], Dr. T. Srinivasa Prasad[3], Dr. Arul Kumar Sengottaiyan[4], Dr. Gayathri Ambedkar[5], Dr. Thodur Madapusi Balaji[6]

[1]Department of Oral Pathology and Microbiology Saveetha Dental College and Hospitals, Saveetha Institute of Medical and Technical Sciences, Saveetha University, Chennai, Tamil Nadu, India

[2]Department of Oral Pathology and, Sri Venkateswara Dental College Thalambur, Chennai Affiliated to the Tamilnadu Dr. MGR Medical University, Chennai, Tamil Nadu, India

[3]Department of Oral and Maxillofacial Surgery, Meenakshi Ammal Dental College and Hospital, Meenakshi Academy of Higher Education and Research (Deemed to be University), Chennai.

[4]Department of Prosthodontics, Department of Prosthodontics, Crown and Bridge University, Vivekananda Dental College for Women, Tiruchengode

[5]Department of Conservative Dentistry and Endodontics, Ragas Dental College and Hospital, Chennai, Tamil Nadu, India

[6]Adjunct Professor Research, Tagore Dental College and Hospital, Near Vandalur, Melakkottaiyur Post, Rathinamangalam, Tamil Nadu, India

Abstract

Unicystic ameloblastoma a rare variant of ameloblastoma accounting for 6 % of ameloblastomas is characterized by slow growth and is less aggressive in comparison with ameloblastoma. It is commonly associated with an impacted third molar. However, under rare circumstances it may be associated with impacted mandibular canine. We present a case of unicystic ameloblastoma in a 31-year-old female patient associated with an impacted mandibular canine. Following treatment, the patient's sister underwent routine radiographic examination which revealed impacted tooth no 43, a finding exactly similar to her sister's case and a small radiolucency surrounding the crown of tooth no 43 which was surgically extracted to prevent further complications. Both the patients are currently under follow-up. With the available clinical data, we hypothesize that the occurrence of jaw lesions associated with impacted teeth could be familial. The present cases highlight the importance of routine radiographic examination and follows up and to obtain family history and screen the family members of patients with impacted teeth to prevent potential complications.

Keywords: *Impacted Mandibular Canine, Ki 67, PCNA, Unicystic Ameloblastoma.*

Introduction

It is a well-known fact that several jaw lesions are associated with impacted teeth. Hence routine radiographic findings associated with impacted teeth should not be ignored. A regular follow up of patients with impacted teeth is essential for assessment of potential complications such as dentigerous cyst, unicystic ameloblastoma.

Ameloblastoma is a benign epithelial odontogenic tumor that accounts for 1% of tumors of odontogenic origin derived from epithelial remnants of the dental lamina and can occur as solid, peripheral, and unicystic

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****Corresponding Author: vsaranya87@gmail.com***

variants. Among the different types of ameloblastoma, unicystic is a rare variant and accounts for 6% of ameloblastomas [1, 2].

Unicystic ameloblastoma was first described by Robinson and Martiney in 1977 [3]. The clinical and radiographic presentation is similar to that of jaw cysts and often warrants histopathological diagnosis. It is more common in the second decade of life with a male predilection [4]. It is most often associated with an impacted third molar in the mandibular region [5]. However, under rare circumstances it has been associated with impacted maxillary canine [6].

Clinical features include swelling, occasional pain, and rarely numbness, when there is a secondary infection discharge occurs. Radiographically the lesion presents as a unilocular well-defined radiolucency. However, in some cases, multilocular radiolucency has been reported. Histopathologic features depict ameloblastomatous epithelium lining cyst with or without mural and luminal invasion. [7]**.** The incidence of unicystic amelobalstoma has been reported to be fifteen percent. Among the histological variant mural type is more common [8]. The disease has a better prognosis in comparison with solid ameloblastoma [9].

Considering the genetic mutations associated with ameloblastoma Heikinheimo et al have reported that BRAF V 600E was most common in the three histological subtypes of unicystic amelobalstoma . Also, the mutation was more common unicystic ameloblastoma than conventional type. Hence the authors concluded that the MAP Kinase pathway was dysregulated that lead to the pathogenesis of unicystic ameloblastoma [10].

With the above data we could hypothesize that unicystic ameloblastoma could be familial owing to the associated genetic mutations mentioned by authors. To report the hypothesis that the occurrence of jaw lesions associated with impacted teeth could be familial by reporting a case of unicystic ameloblastoma in a 31-year-old female patient associated with an impacted mandibular canine and similar small radiolucent lesion associated with impacted 43 in patient's sister.

Case Description

Methods

The below section will describe in detail about the two patients

Patient 1

History

A thirty-one-year-old female patient reported with a chief complaint of swelling in the left lower premolar tooth region for the past two months. Two years ago, the patient had a routine examination for missing permanent lower right canine tooth. On radiographic examination, a 1mm radiolucency surrounding the crown of impacted 43 was seen (Figure 1 A). The patient refused to undergo treatment. Two years later the patient developed swelling in the same region which was slow in growth and was associated with pain spreading from the lower right posterior region to the lower left posterior region.

Clinical Examination

Extraoral examination revealed facial asymmetry and a 7 cm * 3 cm swelling in the mandibular region spanning from tooth number 45 to tooth number 35 extending 15 mm from the angle of the mandible and 5 mm from the lower border of the mandible Skin over the swelling was smooth, the swelling was hard and not associated with pus discharge., non-tender. Orthopantomogram was done.

Provisional Diagnosis

With the available clinical data provisional diagnosis of the dentigerous cyst was given.

Surgery

Under general anesthesia, a mucoperiosteal flap was raised and complete enucleation of the cystic lesion was performed. Impacted tooth and teeth with root resorption were extracted. Chemical cauterization with Carnoy's solution was performed, hemostasis was achieved and 30 black silk sutures were given (Figure 1 C and D).

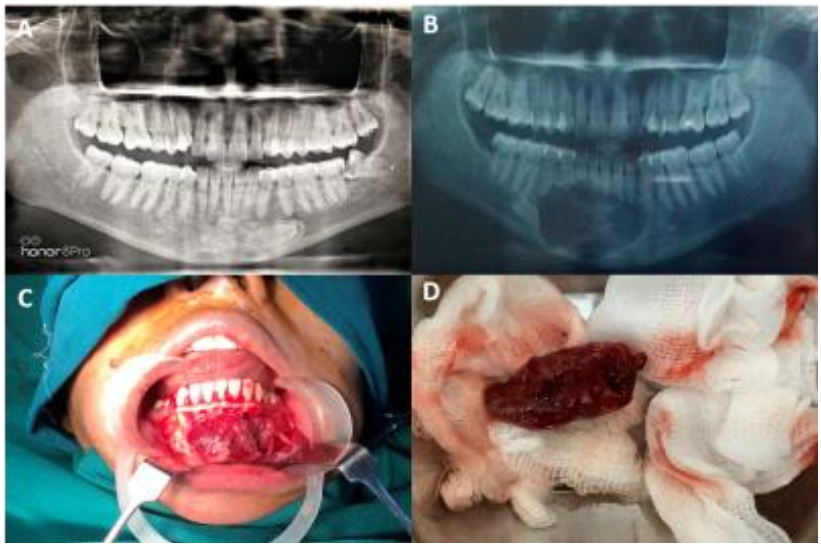

Figure 1. Photographs Depicting the Clinical and Radiographic Image of the 31 Year Old Female Patient

(A) Orthopantomogram of 31 year old female patient depicting the initial size of the lesion 1mm radiolucency surrounding the crown of impacted 43. (B) Orthopantomogram depicting unilocular well-defined radiolucency extending from 35 to 46, associated with impacted 43 and root resorption of the associated teeth. (C and D) Intraoperative images.

Grossing

Two bits of soft tissue was received the larger one measuring 2*1 cm and the smaller bit measuring 0.5 * 0.2 cm, were greyish white, firm inconsistency. The larger bit was grossed into 2 smaller bits for further histopathologic examination. The samples were fixed with 10% neutral buffered formalin and were paraffin embedded according to standard protocol.

Staining

Hematoxylin and Eosin staining

The sections were deparaffinized and rehydrated. Washing of slides was done with distilled wated and stained with Mayers Hematoxylin for sixty seconds and then rinsed with distilled water. Then the specimen was decolourised with 0.5% acid alcohol and then subjected to washing with distilled water. Following this counterstaining was done with eosin for a minute. Following this dehydration was done with gradations of alcohol and clearing was done with three changes of xylene. The slide was dried and mounted with DPX and a coverslip. The slides were viewed under a microscope.

Immunohistochemistry

The formalin fixed paraffin embedded sections were deparaffinized on a hot plate. Antigen retrieval was done using a pressure cooker and cooling of samples were done. Blocking was done with 0.5% bovine serum albumin followed by peroxide and biotin blocking with manufacturer provided blocking agent. The specimens were washed with PBS and addition of Anti CK9, Ki 67, and p53 primary followed by secondary antibodies was done and incubated according to

manufacturer's protocol. This was followed by Mayer's hematoxylin as a counterstain with a 1:10 dilution. The slides were dried and mounted with DPX and cover slip and examined under a microscope.

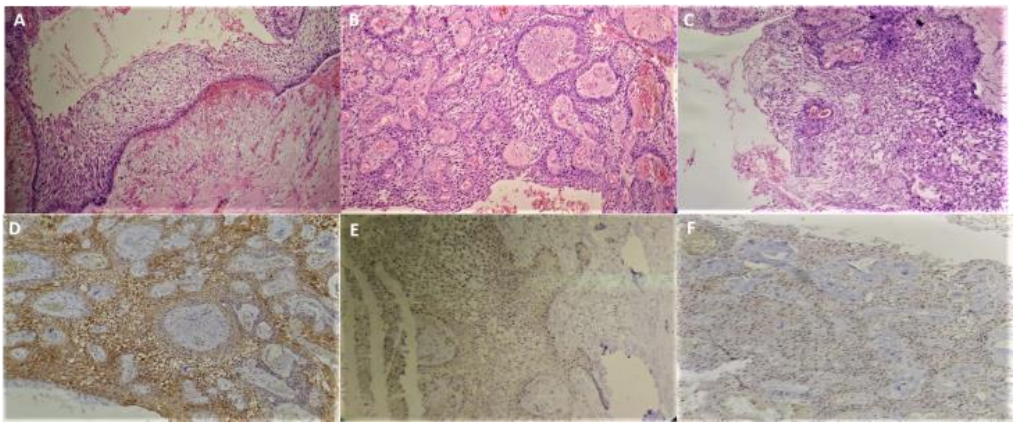

Figure 2. Histopathologic Images of the Received Specimen with Respective Stain

(A to C) H and E section revealing cystic lumen lined by epithelium showing cytomorphometric features of ameloblastoma with mural and intraluminal invasion (magnification 40x). (D) Immunohistochemistry revealing positive stain for cytokeratin. (E) Immunohistochemistry revealing faint positivity for Ki 67. (F) Immunohistochemistry revealing faint positivity for p53.

Patient 2

History

Following treatment, of the first patient, the patient's 28 year old sister visited the dental clinic for a routine dental examination. She did not present with any symptoms.

Radiography

She was subjected to routine radiographic examination and abnormality around 43 was observed.

Surgery:

Surgical extraction of tooth no 43 was done to prevent any associated complications and sequelae.

Histopathology

The obtained tissue sample could not be processed for histopathologic examination on account of the COVID crisis.

Results

Patient 1

Radiography

Orthopantomogram revealed a unilocular well-defined radiolucency extending from 35 to 46, associated with impacted 43, and root resorption of the associated teeth was evident (Figure 1 B).

Histopathology

Microscopic features revealed cystic lumen lined by epithelium showing cytomorphometric features of ameloblastoma comprising of a basal layer of tall columnar cells with reversal of polarity and palisaded nuclei. A thin layer of stellate reticulum-like cells was seen along with intraluminal and mural invasion. Mural invasive islands showed both follicular and plexiform pattern of ameloblastoma (Figure 2 A, B, and C).

Cytokeratin 19 was positive thorough-out the epithelium and stellate reticulum-like cells (Figure 2 D). The cells showed faint positivity for Ki 67 (Figure 2 E) and p53 (Figure 2 F).

With the clinical, radiographic, and histopathologic features a final diagnosis of Ameloblastoma was given.

Follow Up

The patient is under follow up till date and no abnormalities have been detected.

Patient 2

Radiography

OPG revealed impacted tooth no 43, a finding exactly similar to her sister's case and a small radiolucency surrounding the crown of tooth no 43 (Figure 3).

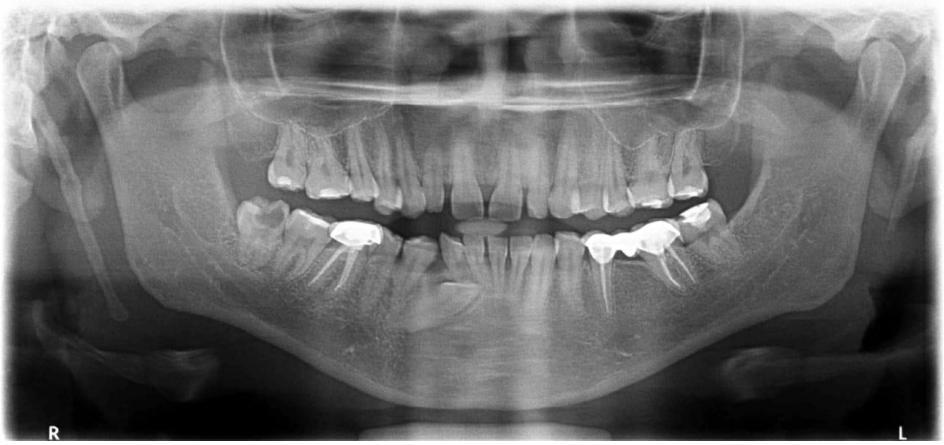

Figure 3. Radiographic Image of the 28 Year Old Female Patient

Orthopantomogram of a 28– year old female patient (sibling of 31 year old patient) depicting small radiolucency surrounding the crown of impacted 43.

Follow Up

The patient is under follow up till date and no abnormalities have been detected.

Discussion

Unicystic ameloblastoma, a rare variant of ameloblastoma accounts for 6 % of ameloblastomas. It occurs in the younger age group with a male predilection [1]. About 50 to 80% of the cases are associated with impacted teeth and the mandibular third molar is more commonly involved, followed by the parasymphysis region, anterior region of maxilla, and posterior region of the maxilla. It has also been noted that the majority of unicystic ameloblastomas are associated with dentigerous cysts and that all such cases occurred in patients less than 30 years of age [11].

In the present case was a 31-year-old female patient was diagnosed with the condition which is concurrent with the literature. It was associated with an impacted mandibular canine. So far very few cases of unicystic ameloblastoma have been reported with impacted canine [12].

The lesion clinically presents as localized swelling associated with facial asymmetry and the rare occurrence of pain. Small lesions are however diagnosed only on routine radiographic examination. In the present case on radiographic examination, a small radiolucency around the impacted tooth was seen when the patient was asymptomatic [9]. However, the patient had ignored the symptom for further treatment that led to the present stage. Radiography revealed unilocular radiolucency associated with root resorption of many teeth that warranted extraction.

Histopathologic criteria for diagnosis are the presence of cystic lumen with ameloblastomatous epithelial lining and a thin layer of stellate reticulum-like cells, mural and luminal invasion may or may not be present.

Ackermann has classified unicystic ameloblastoma into three histologic groups as follows [8].

Group I—luminal unicystic ameloblastoma.

Group II—intraluminal/plexiformunicystic ameloblastoma.

Group III—mural unicystic ameloblastoma. Philipsen and Reichart have classified unicystic ameloblastoma into the following: subgroups as Subgroup 1, Subgroup 1.2, Subgroup 1.2.3 ad subgroup 1.3.[9, 13,14] Based on the above classifications the present case study belongs to Group III and subgroup 1.2.3.

Thus, a definitive diagnosis can be done only an incomplete examination of the entire lesion as preoperative incisional biopsy may not be representative of the entire lesion and may lead to incorrect classification [1]. The exact pathogenesis of unicystic ameloblastoma is unclear. Some authors claim that the lesion arises from a preexisting dentigerous cyst that has transformed while others claim that the lesion arises de novo.

The proposed mechanisms for the development of unicystic ameloblastoma were given by Leider et al. (1985) include an ameloblastic transformation of reduced enamel epithelium of a developing tooth followed by cystic development, those that arise from a dentigerous cyst and cystic degeneration of solid ameloblastomas [15].

The current diagnostic criteria for unicystic ameloblastoma as described by is unaided histologic features showing a single cystic lumen lined by odontogenic epithelium showing features of ameloblastoma. However, Immunohistochemical markers like lectins, Ki-67, PCNA (proliferating cell nuclear antigen (PCNA)), cytokeratins may aid in diagnosis and prognosis. In the present case, Imuunohistochemistry showed positivity for cytokeratin 19 staining thorough-out the epithelium and faint positivity for Ki 67 and p53 which could attribute to a better prognosis of the condition on conservative management. This is concurrent with Li et al who demonstrated Ki 67 positivity and PCNA positivity was associated with recurrence [16].

Complete enucleation, chemical cauterization, and extraction of the teeth with root resorption were performed under General Anesthesia.

The recurrence rate of the lesion is 10 to 20% which is less in comparison with solid ameloblastomas that have a recurrence rate of 50-80% following curettage of multicystic ameloblastomas. Following enucleation alone, a recurrence rate of 30.5% has been reported. The recurrence rate following enucleation with chemical cauterization of 16%, the following marsupialization, and enucleation of 18% and 3.6 % following resection was reported. [17]. Li et al reported that recurrence time interval is up to seven years. [18] Considering histological subtypes the mural variant has more incidence of recurrence. [19]. Both the patients are under follow up for the past 4 years and no recurrence has been reported. This is concurrent with similar cases of unicystic ameloblastoma reported in mandibular region. [20-22].

With the available clinical data, from the two cases we hypothesize that the occurrence of jaw lesions associated with impacted teeth could be familial as this is the first case to have reported similar radiolucenies associated with the same impacted teeth in siblings. The patient’s sister could have developed an odontogenic tumor arising from the remnants of the tooth germ associated with impacted mandibular canine if left without extraction. Mutations associated with the BRAF gene have been implicated in the pathogenesis of ameloblastomas [10, 23]. It is noteworthy that BRAF gene mutations are found to cluster amongst siblings predisposing to various genetic disorders like cardiofaciocutaneous syndrome (CFC) [24]. Whether the BRAF mutation occurrence in siblings could also predispose them to ameloblastoma is an intriguing question although remaining largely unanswered.

Similar locations of impacted teeth have been reported in monozygotic twins

suggesting the genetic predisposition for impacted teeth.[25]. This trend is comparable to our case even though the sisters were not twins. Hence clinicians need to obtain a thorough family history and screen the family members of patients with impacted teeth to prevent potential complications.

Conclusion

The present case highlights the fact that although the lesion has a rare occurrence, patients with impacted teeth should be on regular follow-up for early diagnosis and treatment. Also, the clinicians should obtain a through family history and screen the family members of patients with impacted teeth to prevent potential complications.

Conflict of Interest

None

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None

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