Dysplastic Papilliferous Basaloid Ameloblastoma: Report of a Case

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Abstract

Papilliferous ameloblastoma is an uncommon histologic variant of ameloblastoma. About five cases have been reported in the English literature till date. All five showed squamous differentiation with keratin formation and were diagnosed as papilliferous keratoameloblastoma. The present lesion shows basaloid differentiation and mild dysplasia in addition to papilliferous projections, thus termed dysplastic papilliferous basaloid ameloblastoma.

Ameloblastoma, the most common odostoma has not ceased to intrigue pathologists with its diverse histomorphological patterns. Therefore, we present this uncommon lesion in a 50 year old man with right mandibular swelling.

Key words: Papilliferous, Basaloid, Ameloblastoma, Dysplastic, Non-healing socket.

Introduction

Ameloblastoma was identified by Cusack in 1827, described in 1879 by Falkson and named ameloblastoma by Ivy and Churchill in 1930[1]. Since then, ameloblastoma has been extensively studied and widely reported. However, it remains an enigma till date, with all its variants probably yet to be described and at times challenging to diagnose. This had led to neoplasms having histologic features not classically described in ameloblastoma being so labelled. For example, calcified tissue has been reported within the stroma of a case, while ameloblastoma is classically described as not being differentiated to the point of hard tissue formation [1,2]. In addition, some lesions with no histologic feature of ameloblastoma have been so labelled[1,3,4].

Ameloblastoma, the most clinically significant benign odontogenic neoplasm continues to intrigue pathologists and pose diagnostic challenge due to its myriad of histologic variants, most of which are uncommon. Recognized histological variants include acanthomatous, granular cell, desmoplastic, basal cell, keratoameloblastoma, papilliferous kerato-ameloblastoma and clear-cell ameloblastoma[5]. In this article, we report a very rare variant of ameloblastoma; the papilliferous basaloid ameloblastoma, with dysplastic features in a 50-year-old man.

Case presentation

A 50-year-old man presented at the dental clinic with a three-year history of right mandibular swelling that was preceded by extraction of a painful, right 3rd molar two years before the onset of mandibular swelling. Following the extraction, the socket failed to heal. He was subsequently managed at two different clinics with various antibiotics and analgesics, nonetheless healing was not achieved. In addition, he noticed a jaw swelling in relation to the non healing socket, which was slowly increasing in size and associated with occasional episodes of spontaneous intra-oral bleeding. Five years after onset of symptoms, he presented at the centre where medical history revealed a hypertensive patient on medication, while there was no history of alcohol, tobacco or cola nut consumption. Extra-oral examination revealed an ovoid right mandibular swelling, measuring about 6.5 x 5.0 cm in diameter. The swelling extended from the mandibular angle to the body. Submandibular lymph nodes on the right side could not be palpated because of the tense swelling; however, lymph nodes on the left were palpable, freely mobile and non tender.

Intraoral examination revealed a buccolingual expansion of the mandible extending from 43 to the retromolar area. Teeth related to the swelling were mobile, while an erythematous ulcer with irregular

edges was seen in the retromolar area involving the extraction socket of 48 and extending to the gingivae around 47. Radiographic examination revealed a multilocular radiolucency extending from 43 to the right retromolar area, the teeth in relation to it were displaced.

A clinical impression of primary intra-alveolar carcinoma was made due to the presence of an ulcer, history of occasional spontaneous bleeding and painful 3rd molar whose extraction socket failed to heal. An incisional biopsy was done under local anesthesia. Microscopic examination revealed islands and interconnecting trabeculae of odontogenic cells with a periphery composed of tall columnar ameloblast-like cells showing reversed polarity of the nuclei. The centre consisted of stellate and spindle cells, most of which had undergone basaloid metaplasia, the basaloid cells were interspersed by structures reminiscent of rosettes in a sparsely cellular stroma. Few microcysts were seen, but squamous metaplasia and keratin were absent. Dysplasia, characterized by hyperplasia of the basal layer, was seen. No mitotic figures or areas of necrosis were evident. A diagnosis of dysplastic papilliferous basaloid ameloblastoma was made. The histological features were as shown in Figures 1 and 2.

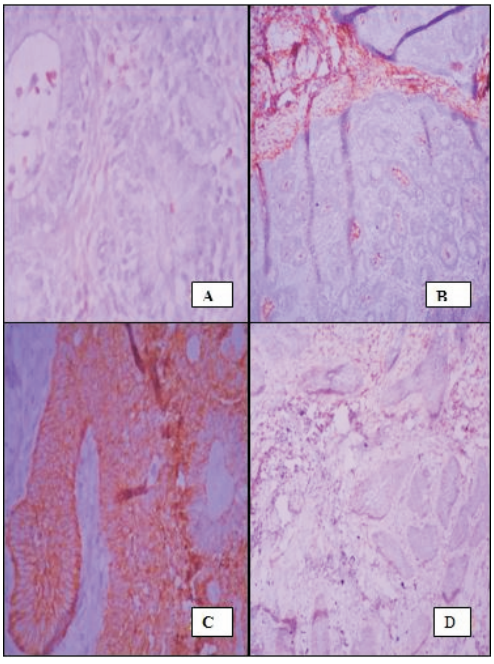

Figure 1: Photomicrographs A (X100), B (X40), C&D (X400) showing several rosettes and papillary extension of odontogenic cells with a fibrovascular core and a periphery of columnar cells (hematoxylin and eosin).

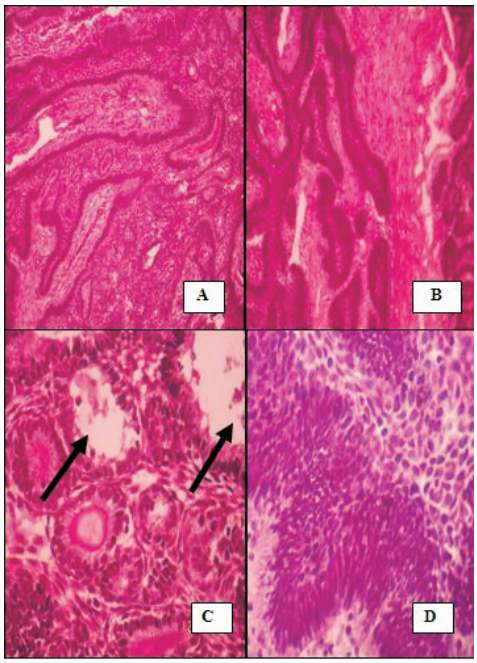

Figure 2: Photomicrographs showing interconnecting trabeculae of odontogenic cells (A: hematoxylin and eosin X100; B: hematoxylin and eosin, X40; C: hematoxylin and eosin x100 with arrows showing micro cysts & D: hematoxylin and eosin x400 showing dysplasia).

The tumour stained strongly for cytokeratin AE1/AE3 and negatively for vimentin, S-100 and NSE as shown in Figure 3.

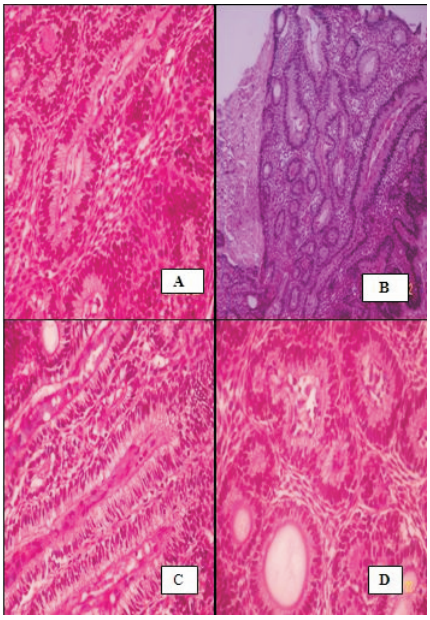

Figure 3: Photomicrographs showing the reaction of papillary basaloid ameloblastoma to immunohistochemical stains (A: Vimentin x100; B: S-100 x100; C: Pancytokeratin 4 x100; D: Neurone specific enolase x100).

Papilliferous ameloblastoma is one of the least reported variants of ameloblastoma; Table 1 is a summary of reports of similar cases [3, 6, 7, 8] **(Table 1).**

Table 1: Clinico-pathologic features of all reported Papilliferous ameloblastoma

Age	Sex	Site	Symptoms	Radiographic features	Features of Ameloblastoma	Cystic cavities	Keratin	Necrosis
57	F	PM	S	ML	None	Yes	Yes	Not indicated
76	F	RPM	U, S	ML	None	Yes	Yes	Yes
26	F	RPM	P, S	ML	Yes	Yes	Yes	Yes
62	M	RPM	DMM	IR	None	Yes	None	Yes
46	M	RPM	S	ML	Yes	Yes	Yes	Yes
50	M	RPM	P, S	ML	Yes	Yes	No	No

F –Female, M – Male, RPM- Right Posterior Mandible, PM-Posterior Mandible, U= Ulcer, S = Swelling, ML= Multilocular Radiolucency, IR-Irregular Radiolucency, DMM-Difficult Mandibular Movement, P = Pain.

Discussion

In 1970, Pindborg described an unusual histological variant of ameloblastoma as papillary keratoameloblastoma. The lesion was diagnosed in a 57-year old female with mandibular mass[7,8]. The second reported case was diagnosed in a 76-year old black woman; the histologic features consisted of cystic cavities most of which were lined by papilliferous odontogenic epithelium, while a few were lined by parakeratinized stratified squamous epithelium. The cystic cavities were filled with necrotic debris and parakeratin. Microscopic features of ameloblastoma were absent in the lesion. The authors attested to the difficulty associated with making a diagnosis and stated that, 'A diagnosis of papilliferous keratoameloblastoma was made after a great deal of uncertainty and difficulty'[3].

The third case was described as an unusual ameloblastoma with cystic follicles containing orthokeratin, parakeratin, desquamated epithelium and necrotic material with dystrophic calcification; in a 26-year old female. The first symptom in the patient was tooth ache, the tooth was extracted but pain persisted, later a jaw swelling was observed in relation to it [6]. This case is very similar to the present case apart from failure of healing in our patient. Histological features consistent with both papilliferous keratoameloblastoma and keratoameloblastoma were seen in the neoplasm [6].

The fourth case was a right mandibular mass in a 62-year-old man. It had cystic cavities lined by epithelium with papillary projections into cystic cavities containing necrotic debris. The lesion showed cribriform, solid and trabecular patterns, as well as focal granular cell differentiation [7]. The authors stated that, "Features resembling keratoameloblastoma or other ameloblastoma variants were not evident". The patient died within six years of diagnosis[7]. The lesion was nonetheless diagnosed as papilliferous keratoameloblastoma. This lesion is unlikely to be an ameloblastoma considering its microscopic features and irregular radiolucent appearance.

The fifth report was a right mandibular mass, in a 46-year-old man. It was a cystic lesion, lined by odontogenic epithelium resembling ameloblastoma with papillary projections into cystic cavities and areas of keratinization [8].

The present lesion is of interest for several reasons. First, the lesion displayed two histological features that are quite rare in ameloblastoma: papilliferous appearance and basaloid differentiation [4,7]. Secondly, there were features of dysplasia in the tumour. In addition, rosette-like structures were seen in areas with basaloid metaplasia. Although the cellular mechanism responsible for rosette formation is not fully known, the 'rosettes' in the index case of ameloblastoma seemed to have resulted from sections through papillary projections of odontogenic cells with peripherally placed ameloblast like cells.

Four main types of rosettes have been described in histopathology: Homer Wright, Flexner Wintersteiner, true ependymoma rosettes and perivascular pseudorosettes. Homer Wright rosettes are characterized by a halo of tumour cells surrounding a centre of neuropils[9,10]. They are more commonly seen in peripheral neuroectodermal tumor (PNET) and medulloblastoma. Flexner Wintersteiner rosettes have a halo of tumour cells and a central core made of

cytoplasmic extension of the cells [9,10]. These rosettes are seen commonly in retinoblastoma, but may be found in pineoblastomas and medulloepitheliomas. True ependymoma rosettes have tumour cells arranged around empty spaces and are found in ependymomas [9,10]. They are believed to be attempts at forming ventricles with ependymal lining. The fourth type of rosettes are perivascular pseudo-rosettes, seen in medulloblastomas, ependymomas, PNETs, central neurocytomas, and less often in glioblastomas. The tumour cells are arranged around blood vessels in perivascular pseudo-rosettes; they are termed pseudo-rosette because the middle structure is not the part of the tumour[9,10]. Typically, rosettes are not part of the microscopic features of ameloblastoma[11].

The rosettes in this report are peri fibrovascular pseudo-rosettes; tumour cells are arranged around the fibro-vascular core of the numerous papillae in the tumour. The clinical implications of this microscopic feature in ameloblastoma is presently unknown. More reports of ameloblastoma with rosette formation may shed more light on effects on clinical behaviour and prognosis. However, in 1998, Weir et al[12], reported a case of ameloblastoma with rosette formation in a 53-year-old woman whose medical history was significant for excision of right mandibular ameloblastoma at ages 8 and 43, as well as lobectomy for a metastasis to the left lower lobe of the lung. At the age of 45 years, a second metastasis was resected from the right middle lobe of the lung. At 53 years of age, she presented with another right mandibular growth and a left lobe nodule, both were essentially similar microscopically. They were diagnosed as recurrent and metastatic ameloblastoma, respectively. The rosettes described by Weir et al, were also peri fibrovascular pseudo rosettes[12].

Tooth extractions are common procedures in dental practice, often done for grossly broken down or very mobile teeth but should be done only when indicated after adequate investigation. This case underscores the importance of proper pre-operative evaluation, in order to ascertain the pathology of symptomatic teeth and institute the most appropriate management. It also emphasizes the need to investigate non healing extraction sockets[13]. In this patient, the extraction socket failed to heal more than one year after extraction. Generally, extraction sockets show appreciable evidence of healing two weeks post extraction and the outline of the socket should be invisible on radiographic examination three to four months post exodontia[14]. However, diverse pathologies ranging from alveolar osteitis, osteomyelitis, presence of foreign body, osteoradio-necrosis to life threatening diseases, such as malignancies have prevented healing of extraction sockets[15]. Thus, non-healing extraction sockets, especially after conventional treatment should be assessed radiologically and microscopically. As Chen and Peron advocated, that the first step in managing a non-healing extraction socket is to rule out an intra-osseous malignancy [16].

Conclusion

In conclusion, we present a case of papilliferous basaloid ameloblastoma with rosette-like structures and emphasize the need to investigate all non-healing extraction sockets, as well as the compulsion of adequate pre-extraction investigation.

References

1. Gupta N, Anjum RM Gupta S, Lone P. Ameloblastoma of the mandible: A case report with review of literature. Int. J Head and Neck Surg 2012; 3 (1):56-58.
2. Takeda Y, Satoh M, Nakamura S, Ohya T. Keratoameloblastoma with unique histological architecture: an undescribed variation of ameloblastoma. Virchows Arch. 2001; 439 (4):593-596.
3. Altini M, Slabbert HD, Johnston T. Papilliferous keratoameloblastoma. J Oral Pathology & Medicine 1991; 20(1):46–48.
4. Shakya H, Khare V, Pardhe N, Mathur E, Chouhan M. Basal cell ameloblastoma of mandible: a rare case report with review. Case Reports in Dentistry 2013. http://dx.doi.org/10.1155/2013/187820. Accessed 22/04/15.
5. Adeyemi BF, Adisa AO, Fasola AO, Akang EEU. Keratoameloblastoma of the mandible. J Oral Maxillofac Pathol. 2010; 14 (2):77-79.
6. Norval EJ, Thompson IO, van Wyk CW. "An unusual variant of keratoameloblastoma," J Oral Pathol 1994; 23(10):465–467.
7. Collini P, Zucchini N, Vessecchia G, Guzzo M. Papilliferous keratoameloblastoma of mandible: a papillary ameloblastic carcinoma: report of a case with a 6-year follow-up and review of the literature. Int J Surg Pathol. 2002; 10 (2):149-155.
8. Mohanty N, Rastogi V, Misra SR, Mohanty S. Papilliferous keratoameloblastoma: an extremely

rare case report. Case Reports in Dentistry 2013. http://dx.doi.org/10.1155/2013/706128. Accessed on 22/04/15.

9. Wippold FJ, Perry A. Neuropathology for the neuroradiologist: rosettes and pseudo-osettes. Am J Neuroradiology 2006; 27:488-492.

10. Das D, Bhattacharjee K, Barthakur SS et al. A new rosette in retinoblastoma.Indian J Ophthalmol. 2014;62(5):638-641.

11. Gardener DG, Heikinheimo K, Shear M, HP Philipsen, Coleman H. Ameloblastoma. In: Barnes L, Eveson JW, Reichart P, Sidransky D. World Health Organization Classification of Tumors. Pathology and Genetics of Tumors of the Head and Neck. Lyon: IARC Press; 2005

12. Weir MM, Centeno BA, Szyfelbein WM. Cytological features of malignant metastatic ameloblastoma: a case report and differential diagnosis. Diagn Cytopathol. 1998;18(2):125-130.

13. Cían J.H, Leo F.A. The non-healing extraction socket: a diagnostic dilemma – case report and discussion. Journal of the Irish Dental Association 2016; 62 (4): 215-220.

14. Haghighat A, Hekmatian E, Abdinian M, Sadeghkhani E. Radiographic Evaluation of Bone Formation and Density Changes after Mandibular Third Molar Extraction: A 6 Month Follow up. Dent Res J (Isfahan). 2011;8(1):1-5.

15. Lorè B, Gargari M, Ventucci E, Cagioli A, Nicolai G, Calabrese L. A complication following tooth extraction: chronic suppurative osteomyelitis. Oral Implantol (Rome). 2013;6(2):43-47.

16. Chen N, Peron JM. The non-healing of the buccal mucosa after tooth extraction. Apropos a case of histiocytosis X. Rev Stomatol Chir Maxillofac. 2000; 101 (1):33-35.