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Introduction

Ameloblastoma is a benign tumor originating from odontogenic tissue, predominantly found in the maxillofacial region, with several histological subtypes. Hybrid ameloblastoma, first described by Waldron and El-Mofty in 1987, exhibits histological characteristics of both desmoplastic and conventional ameloblastoma.[12,]The incidence of hybrid ameloblastoma is 1.1% and most commonly occurs in individuals with a mean age of 24.5 years, with a higher prevalence in females compared to males.[1] This subtype primarily affects the mandible, with a ratio of 5:3 relative to the maxilla.[3]Radiographically, hybrid ameloblastoma typically presents with a mixed radiolucent and radiopaque appearance and often has with irregular borders. In some cases, it may also exhibit multilocular radiolucency similar to conventional ameloblastoma.[4]Histopathologically, hybrid ameloblastoma features both desmoplastic patterns and conventional ameloblastoma patterns, predominantly follicular and plexiform. Although less common, acanthomatous, basal cell, and granular cell patterns may occasionally be observed.[3] In this report, we present a case of hybrid ameloblastoma in a 60-year-old male patient, localized to the right mandibular region.

Case History/ Examination

A 60-year-old male presented to the Department of Oral Medicine and Radiology with a complaint of a painless swelling in the lower right jaw region for the past three months. Examination revealed an extraoral swelling on the lower right side of the face **(Figure 1).** Intraoral examination showed buccal and lingual cortical expansion with an eggshell crackling sensation upon palpation **(Figure 2).**

Methods

Aspiration of the swelling yielded serosanguinous fluid. Radiographic evaluation using an orthopantomogram (OPG) showed a single, localized, well-defined, scalloped, multilocular radiolucency with a corticated border in the right body of the mandible. The lesion extends from the periapical region of tooth 41 to the mesial aspect of tooth 47. Additionally, the root apices of teeth 41, 42, 43, and 44 appear blunt, indicating possible root resorption **(Figure 3).** Computed tomography revealed thinning, expansion, and perforation of both the buccal and lingual cortical plates at multiple sites **(Figure 4).** Based on the comprehensive clinical and radiographic examination, the differential diagnosis included unicystic ameloblastoma, odontogenic keratocyst, and ameloblastic carcinoma.

An incisional biopsy was performed under local anesthesia, and the specimen was preserved in a 10% formalin solution for further histopathological analysis. The macroscopic examination revealed a tissue specimen that varied in color from greyish white to light brown to dark brown, firm in consistency, and measured 3.5 cm by 0.6 cm**(Figure 5).** Histopathological examination using routine hematoxylin and eosin (H&E) staining revealed connective tissue stroma containing anastomosing cords, strands, nests, sheets, and islands of odontogenic epithelium **(Figure 6A).** The islands featured peripheral tall columnar cells with hyperchromatic, round nuclei, displaying a reversal of polarity reminiscent of ameloblastoma-like cells. The central areas within these islands contained loosely connected, angulated cells resembling stellate reticulum-like

cells**(Figure 6B).** Some of the islands exhibited bizarre shapes and were surrounded by areas of hyalinization, resembling features of desmoplastic ameloblastoma **(Figure 6C).** Additionally, cystic degeneration and squamous metaplasia were observed within some of the tumor islands **(Figure 6D).** The surrounding stroma contained mild inflammatory cell infiltrate, moderate vascularity, areas of hemorrhage, and bony trabeculae.

Based on the correlation of clinical and histopathological features, a diagnosis of hybrid ameloblastoma (desmoplastic ameloblastoma with follicular ameloblastoma) was established.

not-yet-known not-yet-known

not-yet-known

unknown

Conclusion and Results Following the final diagnosis, marsupialization of the lesion was performed, followed by dredging. The patient was monitored closely during follow-up visits every two weeks for 6 months. At the six-month postoperative follow-up, the patient remained free of any signs of disease recurrence.

PATIENT PERSPECTIVE

Undergoing surgery and regular follow-up has been challenging, but the patient is committed to the treatment plan, understanding the importance of close monitoring to prevent recurrence. The support from healthcare professionals has been crucial in navigating the physical and emotional aspects of this diagnosis.

Conclusion

Hybrid ameloblastoma is a rare variant characterized by diverse clinical, radiographic, and histopathological features. The clinical significance of its desmoplastic and conventional components is unclear, necessitating further research. Accurate radiographic evaluation and thorough histopathological analysis are essential for proper diagnosis, and vigilant long-term monitoring is crucial to manage recurrence risk.

AUTHORSHIP CONTRIBUTION

Nature of Work	1A	2A	3A	4A
Concept	\tightlist	\tightlist		
Design	\tightlist	\tightlist	\tightlist	\tightlist
Definition of intellectual content		\tightlist		\tightlist
Literature search	\tightlist	\tightlist	\tightlist	\tightlist
Manuscript preparation	\tightlist	\tightlist	\tightlist	\tightlist
Manuscript review & editing	\tightlist	\tightlist	\tightlist	\tightlist

REFERENCES

https://doi.org/10.1016/j.ajoms.2019.09.006

1. Sheikh TA, Kamble PP, Deshmukh G, Vermani A, Verma M. A Curious Case of Hybrid Ameloblastoma. Cureus. 2023;15(7).**doi:https://doi.org/10.7759%2Fcureus.42512** 2. Singh R, Gupta M, Chatterjee S, Arora KS. Hybrid ameloblastoma: a mystified histopathological entity. BMJ Case Reports CP. 2019;12(4):e229834.**10.1136/bcr-2019-229834** 3. Nariai Y, Yanai C, Iwahashi T, Kanno T. Clinical features of giant hybrid ameloblastoma of the mandible: A case report with long-term observation of natural growth before treatment. Oral and Maxillofacial Surgery Cases. 2019;5(3):100116.**doi:https://doi.org/10.1016/j.omsc.2019.100116** 4. Rai S, Misra D, Prabhat M, Jain A, Jain P. Hybrid ameloblastoma of anterior maxilla: A rare and puzzling pathologic entity–Case report with systematic review. Contemporary Clinical Dentistry. 2019;10(1):147-53.**doi:10.4103/ccd.ccd_341_18** 5.

Kumar VM, Chakravarthy A, Sathyanarayanan R, Raghu K, Reddy CD. Hybrid ameloblastoma arising from a treated odontogenic keratocyst of the mandible: a case report with literature review. Indian Journal of Otolaryngology and Head & Neck Surgery. 2022:1-9.**doi:https://doi.org/10.1007/s12070-021-02889-y** 6. Reichart PA, Philipsen HP. Odontogenic tumors and allied lesions. 1st ed. London: Quintessence; 2004. 47-9 p.7. Chintamaneni Raja Lakshmi M, Sujana Mulk Bhavana M, Sai Madhavi Nallamilli M, Meka Poorna Venkata Prabhat M, Gummadapu Sarat M, Chennupati Anuradha B. Hybrid ameloblastoma of the maxilla: a puzzling pathology. Iranian Journal of Medical Sciences. 2016;41(4):340. **PMID: 27365557** 8. Karasu B, Özalp Ö, Özbilim G, Sindel A, Altunay B. A Hybrid Ameloblastoma in Posterıor Mandible: A Case Report. Selcuk Dental Journal. 2024;11(1):55-8.**doi:https://doi.org/10.15311/selcukdentj.1237397** 9. Effiom O, Ogundana O, Akinshipo A, Akintoye S. Ameloblastoma: current etiopathological concepts and management. Oral diseases. 2018;24(3):307-16. **doi:https://doi.org/10.1111/odi.12646** 10. Patil SK, Telagi N, Nair MR, Mujib BA. Desmoplastic ameloblastoma: A case report. Journal of Family Medicine and Primary Care. 2020;9(5):2544-7. **doi:10.4103/jfmpc.jfmpc_322_20** 11. Desai PM, Khorate M, Figueiredo N. Desmoplastic ameloblastoma of the mandible: A rare case report. Journal of Indian Academy of Oral Medicine and Radiology. 2020;32(1):81-5.**doi:10.4103/jiaomr.jiaomr_165_19** 12. Iwase M, Fukuoka A, Tanaka Y, Saida N, Onaka E, Bando S, et al. Hybrid desmoplastic/follicular ameloblastoma of the mandible: A case report and review of the literature. Case Reports in Pathology. 2017;2017.**https://doi.org/10.1155/2017/7031414** 13. Kakuguchi W, Ohiro Y, Nakazawa S, Naito R, Moritani Y, Nakamichi Y, et al. Application of the dredging method in a case of recurrent ameloblastoma that had spread over a large region of the mandible. Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology. 2020;32(1):44-8. **doi:** 14. Managutti A, Menat S, Shah D, Katwala H, Managutti S. Scholars Journal of Medical Case Reports.4(9):687-91.**doi:10.36347/sjmcr.2016.v04i09.017** 15. Cortese A, Spirito F, Claudio PP, Lo Muzio L, Ruggiero A, Gargiulo M. Mandibular reconstruction after resection of ameloblastoma by custom-made CAD/CAM mandibular titanium prosthesis: two case reports, finite element analysis and discussion of the technique. Dentistry Journal. 2023;11(4):106. **doi:https://doi.org/10.3390/dj11040106**

Table 1: Radiographic Presentation of Desmoplastic Ameloblastoma

Type	Description	Radiographic Features
Type I (Osteofibrosis type)	Mixed Radiolucent-Radiopaque	Mixed areas of radiolucency and radiopacity with ill-def
Type II (Radiolucent type)	Unilocular Radiolucency	Single large radiolucent lesion with well-defined or ill-de
Type III (Compound type)	Multilocular Radiolucency	Multiple radiolucent compartments with scalloped or ir

Legends

Figure 1: Extraoral picture showing swelling in the right mandibular region.

Figure 2: Intraoral picture showing swelling in the buccal and lingual aspect in relation to the mandibular right canine and premolar.

Figure 3: OPG showing well-defined radiolucent area (white arrows) with corticated border extending from the midline of 41 to the mesial aspect of 47.

Figure 4: Computed tomography (Axial section) showing thinning and perforation of the buccal and lingual cortical plate

Figure 5: Macroscopic appearance

Figure 6 A: Anastomosing cords, strands and islands of odontogenic epithelium in 4x magnification, B: Islands showing peripheral tall columnar cells (black arrow) and central stellate reticulum-like cells (white arrow) in 40x magnification, C: Bizarre shaped island and areas of hyalinization (black arrow) in 10x magnification, D: Islands showing squamous metaplasia (black arrow) in 40x magnification.

Figure 7: Different Imaging modalities in Ameloblastoma

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FIGURES.docx available at https://authorea.com/users/816297/articles/1221888-not-yet-known-not-yet-known-not-yet-known-unknown-unveiling-hybrid-ameloblastoma-a-case-study-of-an-aggressive-variant-with-unique-diagnostic-challenges