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# Journal of Oral Medicine, Oral Surgery, Oral Pathology and Oral Radiology



Journal homepage: http://www.joooo.org

#### Case Report

# Basal cell ameloblastoma in the mandibular symphysis: A unique case report of a rare histological variant with uncommon presentation

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#### Abstract

Basal cell ameloblastoma is an uncommon histological variant of ameloblastoma, characterised by basaloid epithelial cells with minimal peripheral palisading, often mimicking other basaloid neoplasms. We report a case of a 64-year-old male presenting with a five-year history of swelling in the anterior mandible. Radiographic assessment including orthopantomography and computed tomography revealed a well-demarcated intraosseous lesion in the symphyseal region. A definitive diagnosis was established through histopathological examination, confirming basal cell ameloblastoma. Due to its rarity and histopathological overlap with malignancies such as basal cell carcinoma and adenoid cystic carcinoma, accurate diagnosis necessitates comprehensive clinicopathological correlation. Given the limited data on its biological behaviour, long-term follow-up is advised.

Keywords: Basal cell ameloblastoma, Histological variant, Basal cell carcinoma.

Received: 31-05-2025; Accepted: 05-07-2025; Available Online: 29-09-2025

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# 1. Introduction

Odontogenic tumours represent a biologically diverse group of lesions originating from the odontogenic apparatus, exhibiting a broad spectrum of histopathological subtypes and clinical behaviours. Ameloblastoma, although benign, is a locally aggressive epithelial neoplasm that comprises approximately 1% of all odontogenic cysts and tumours affecting the maxillofacial region, and about 10% of all odontogenic tumours.<sup>1</sup>

According to the World Health Organization (WHO), ameloblastoma is defined as a benign, locally invasive, and polymorphic odontogenic epithelial neoplasm, most commonly exhibiting follicular or plexiform architectural patterns within a fibrous stromal background.

A histopathological review by Adebiyi et al. reported the follicular subtype as the most frequent (64.9%), followed by plexiform (13.0%), desmoplastic (5.2%), and acanthomatous

(3.9%) variants. The basal cell variant, an infrequent and poorly characterized subtype, accounted for only 2.6% of cases, underscoring its rarity and the diagnostic complexities associated with its identification.<sup>2</sup> Basal cell ameloblastoma is a rarest histological variant of ameloblastoma, although primarily reported in peripheral locations, intraosseous occurrences are exceptionally rare, with only 30 cases documented in the literature to date.<sup>3</sup> Due to its infrequency, data regarding its clinical behaviour and long-term prognosis remain inconclusive. Histologically, this variant is composed of basophilic basaloid cells arranged primarily in trabecular or nested patterns, with minimal or absent peripheral palisading, which distinguishes it from more conventional ameloblastoma subtypes. Given the paucity of documented cases, establishing its biological aggressiveness remains challenging. Nonetheless, the available evidence suggests that its recurrence potential and risk of malignant

\*Corresponding author: Shadiya C Email: shadiyacmohammed7@gmail.com transformation are comparable to those of other ameloblastoma variants.<sup>4</sup>

One of the significant diagnostic challenges associated with basal cell ameloblastoma lies in its histopathological resemblance to certain malignant epithelial tumors, particularly basaloid squamous cell carcinoma, solid-type adenoid cystic carcinoma, and basal cell carcinoma occurring within the oral cavity. This morphological similarity may lead to misdiagnosis, especially in small or fragmented biopsy specimens. Therefore, a thorough evaluation incorporating clinical, radiographic, and histopathological findings is imperative to achieve an accurate diagnosis.<sup>3</sup>

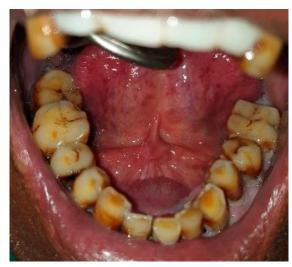
# 2. Case Report

A 64-year-old male patient presented with a chief complaint of swelling in the anterior mandibular region, progressively enlarging over four years. The patient reported mild pain and mobility of teeth in the affected area, along with a history of intermittent tingling and numbness in the surrounding soft tissues for the past six months. There was no relevant medical, dental, or family history, and no history of trauma or discharge.

Intraoral examination showed a solitary, well circumscribed, roughly oval shaped swelling with an irregular surface on the labial aspect of the anterior mandibular region as seen in Figure 1. The lesion extended mediolaterally from the distal surface of tooth 43 to 35 and superoinferiorly from the attached gingiva to the labial and buccal vestibules, involving the region corresponding to teeth 43 through 35. There was obliteration of the labial and buccal vestibules from teeth 43 to 35, with Eggshell crackling suggesting significant cortex expansion. Grade II mobility was observed in teeth 31, 32, and 41. Additionally, a welldefined, firm, smooth, oval swelling measuring  $1 \times 1$  cm was present on the lingual aspect near the floor of the mouth in mandibular the anterior region. (Figure 2) Orthopantomograph as observed in Figure 3 revealed.



**Figure 1:** Intraoral clinical photograph showing well circumscribed non smooth swelling present in the labial aspect of mandibular anterior region



**Figure 2:** Intraoral clinical photograph showing oval shaped smooth swelling in the lingual aspect of mandibular anterior region near the floor of mouth



**Figure 3:** OPG showing multilocular radiolucency with scalloped borders present on the mandibular anterior region crossing the midline

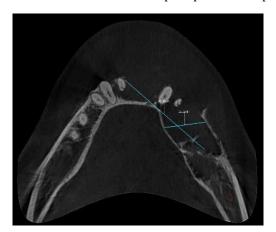
A multilocular radiolucent lesion in the anterior mandibular region, extending from tooth 43 to 36. The lesion appeared roughly oval in shape with scalloped margins. Notable effects on surrounding structures included knifeedge root resorption of teeth 41 and 31, displacement of teeth 43, 42, 33, and 35 without evidence of root resorption, and inferior displacement of the left inferior alveolar nerve canal. Axial section of CBCT (Figure 4) revealed an expansile hypodense lesion involving the mandibular midline, with evident resorption and perforation of the labial cortical plate. The lesion extended from the mesial aspect of tooth 43 to the distal aspect of tooth 36. Sagittal CBCT section (Figure 5) revealed significant buccal and lingual cortical expansion with evident perforation of the buccal cortical plate. Knifeedge root resorption was observed in relation to tooth 31. Cross-sectional CBCT view (Figure 6) revealed marked cortical thinning accompanied by perforation and erosion of both the buccal and lingual cortical plates, consistent with an expansile and potentially aggressive intraosseous lesion.

To establish a definitive diagnosis, an incisional biopsy was performed. Hematoxylin and eosin-stained sections

revealed lesional tissue composed of multiple nests and islands of uniform basaloid cells arranged in a lobular configuration, separated by fibrous connective tissue septa. The central areas of the epithelial nests lacked stellate reticulum-like cells. Peripheral cells were cuboidal to short columnar in shape and exhibited reversal of nuclear polarity.

These histological features were consistent with a diagnosis of basal cell ameloblastoma.

The patient was advised to undergo definitive surgical management and is currently under regular clinical and radiological follow-up to monitor for any signs of recurrence or postoperative complications.



**Figure 4:** CBCT axial section showing expansile hypodense lesion involving the mandibular midline with resorption & perforation of labial cortical plate

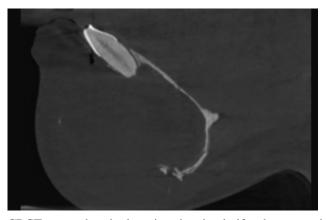
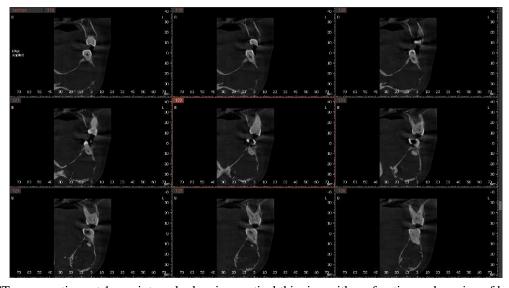


Figure 5: CBCT cropped sagittal section showing knife edge resorption wrt 31



**Figure 6**: CBCT cross sections at 1 mm intervals showing cortical thinning with perforation and erosion of bucca and lingual cortex

### 3. Discussion

Ameloblastoma is a benign but locally aggressive odontogenic tumor, comprising approximately 1% of all neoplasms and cystic lesions affecting the jaws. It demonstrates a broad spectrum of histopathological patterns and clinical behaviors. The basal cell variant of ameloblastoma (BCA) is an infrequent histological subtype, reported to constitute less than 2% of all ameloblastomas. It is characterized by its histological similarity to basal cell carcinoma (BCC) of the skin and other basaloid neoplasms, which often complicates the diagnostic process.<sup>5</sup>

In the present case, we document a 64-year-old male presenting with an asymptomatic swelling localized to the anterior mandibular region, extending from tooth 43 to 36. This anatomical location is notably uncommon for both conventional and basal cell variants, which predominantly manifest in the posterior mandible, particularly the molar and ramus areas. Furthermore, the age of the patient lies outside the typical range cited in the literature, which reports the highest incidence of BCA between the third and fourth decades of life. These deviations highlights the rarity of our case in terms of both anatomical site and age of presentation.

Radiographic evaluation using orthopantomography and cone-beam computed tomography revealed a well-demarcated multilocular radiolucent lesion, accompanied by root resorption, expansion of buccal and lingual cortical plates, and cortical perforation. These imaging features are in accordance with those described in previously published cases of intraosseous ameloblastomas, including the study by Sridhar et al.,<sup>6</sup> though their case involved the posterior mandible and younger age group. The radiographic characteristics reflect the tumor's potential for significant local bone destruction, despite its benign histological nature.

Histopathological analysis of the biopsy specimen demonstrated islands of uniform basaloid epithelial cells without central stellate reticulum-like differentiation. The peripheral cells were cuboidal to columnar in morphology, exhibiting nuclear palisading and reverse polarity, and the nests were separated by delicate fibrous septa, creating a lobular architecture. These findings are consistent with the diagnostic criteria for BCA.<sup>7</sup> It is important to note that certain histological features of BCA can mimic malignant basaloid neoplasms such as basaloid squamous cell carcinoma (BSCC), solid variant of adenoid cystic carcinoma (ACC), and intraoral BCC. Distinguishing these entities is essential due to their differing prognostic implications and management protocols.

Although immunohistochemical (IHC) analysis was not performed in this case, literature suggests that it plays a vital role in differential diagnosis when histological ambiguity exists. Markers such as cytokeratins AE1/AE3, MNF116, and the absence of Ber-EP4 and CD117 (c-kit) expression support the diagnosis of BCA, as opposed to ACC or BCC, which

show distinct immunoprofiles.<sup>8</sup> Additionally, proliferative indices such as PCNA and Ki-67, which have been shown to be elevated in the basal cell variant compared to other ameloblastoma subtypes, may aid in evaluating the biological activity of the lesion.<sup>9,10</sup>

The therapeutic approach to ameloblastomas, including their basal cell variant, remains a subject of clinical discussion. Options range from conservative methods such as enucleation and curettage to radical resection. Recurrence rates have been notably higher following conservative treatment, with reported rates ranging from 55% to 90%. Radical surgical excision with adequate bony margins remains the preferred method to minimize recurrence risk. In the present case, surgical resection with planned margins was advised, and the patient was placed under periodic follow-up for early detection of any recurrence, in accordance with standard guidelines. 12

Given the limited number of reported cases, the prognosis of BCA remains unclear. However, the tumor is generally considered to behave similarly to other solid/multicystic ameloblastomas in terms of local aggressiveness and recurrence potential. Long-term surveillance is essential due to the possibility of late recurrence and the histological resemblance of BCA to malignancies, which may lead to diagnostic delays if not thoroughly assessed.<sup>13</sup>

This case illustrates the critical importance of correlating clinical, radiographic, and histopathological findings to establish a precise diagnosis. It also emphasizes the need for heightened awareness of unusual presentations of BCA, particularly in older patients and atypical anatomical locations, to facilitate early and appropriate intervention.

## 4. Source of Funding

None.

# 5. Conflict of Interest

None.

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Cite this article: Shadiya C, Aggarwal A, Asdullah M, Verma P. Basal cell ameloblastoma in the mandibular symphysis: A unique case report of a rare histological variant with uncommon presentation. *J Oral Med Oral Surg Oral Pathol Oral Radiol*. 2025;11(3):129–133.