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Case Report

Mandibular Unicystic Ameloblastoma: Case Report with 23 Years of Follow-Up

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Abstract

Unicystic ameloblastoma (UA) is a distinct clinicopathological variant of ameloblastoma that predominantly affects young patients and exhibits a wide range of histopathological patterns. The aim of this case report is to emphasize the importance of histopathological diagnosis and to present immediate mandibular reconstruction using a microvascular free fibula flap, with a 23-year follow-up. A 25-year-old man presented to the emergency department with a 24-hour history of pain and severe swelling in the left mandibular region that did not respond to painkillers. Clinical examination revealed swelling in the left retromolar area, tenderness on palpation, and absence of the left mandibular third molar. The patient reported that no third molar had ever been extracted. Periapical radiography revealed a large osteolytic lesion with displacement of the impacted third molar toward the inferior border of the mandible. Panoramic radiography and computed tomography confirmed an extensive osteolytic lesion involving the entire left mandibular ramus up to the condylar region. An incisional biopsy was performed, and histopathological examination confirmed unicystic ameloblastoma with mural nodules. The patient underwent left hemimandibulectomy followed by immediate reconstruction using a microvascular free fibula flap.

Keywords: *ameloblastoma, odontogenic tumors, jaw neoplasms, unicystic ameloblastoma*

Introduction

In most literature, intraosseous odontogenic lesions are detected through routine radiographic studies or in consultations prompted by pain or swelling. In such context, presumptive diagnoses include dentigerous cyst, radicular cyst, or keratocyst. However, ameloblastoma, both in its conventional and unicystic form, must be considered in the differential diagnosis.

An ameloblastoma is a benign, locally aggressive, epithelial odontogenic tumour accounting for around 10% to 13% of all odontogenic tumours. It is slow-growing, and it usually does not display any clinical signs until the lesion reaches sensitive structures and causes root resorption, displacement of the inferior alveolar nerve canal, or tooth displacement. The third molar region in the lower mandibular ramus is one of the most frequently affected areas (1).

Conventional ameloblastoma usually affects people during their third and fourth decades of life. On the other hand, unicystic ameloblastoma affects mainly young people, with a peak incidence around their twenties and with no sex predilection (2).

Unicystic ameloblastoma exhibits distinct histological features according to the growth subtypes described in the literature. Treatment must be planned considering the size of the lesion, radiographic appearance, adjacent structures involvement and histological pattern, since the latter determines the biological behaviour of the lesion (3).

During an emergency consultation prompted by an inflammatory condition, a unicystic ameloblastoma was detected. Due to the size of the lesion, it was necessary to perform a microvascular fibula free flap reconstructive surgery. The patient has had a 23-year follow-up.

Case Presentation

In 2003, a 25-year-old male patient with no significant medical records attended an emergency consultation at the Maxillofacial Surgery Department.

The chief complaint was a 24-hour history of pain in the lower left jaw that did not respond to painkillers. Clinical examination revealed a tender oedema in the left retromolar trigone area; and absence of unerupted tooth #38. An intraoral periapical radiograph was performed, which revealed an osteolytic lesion with displacement of tooth #38 towards the basal bone (Figure 1). A panoramic radiograph and a maxillofacial computed tomography were requested to complete the examination, along with laboratory tests to schedule an incisional biopsy.

Laboratory results were within normal limits. The panoramic radiograph revealed an extensive, unilocular osteolytic lesion related to the impacted tooth #38 that was displaced towards the mandibular basal bone. The lesion involved the left lower mandibular ramus up to the condylar neck and produced expansion of the ramus and the coronoid process, along with cortical thinning (Figure 2). The computed tomography confirmed expansion and thinning of all the mandibular cortices in the left ramus area from tooth #37 with displacement of tooth #38 towards the basal area; the lesion extended up to the condylar neck, preserving only its head (Figures 3 and 4).

An incisional biopsy was performed, the specimen was fixed in formalin for histopathological examination, and results confirmed a luminal unicystic ameloblastoma with mural growth. Immunostaining was conducted to mark the tumour epithelium CK5 and CK18, both of which were positive (Figures 5 and 6). The patient was offered the following treatment: a left hemimandibulectomy, including the condyle, alongside immediate reconstruction using a microvascular fibula free flap.

The patient underwent surgery, during which a hemimandibulectomy was performed at the mesial resection margin of tooth #36, including the condyle. To reconstruct the surgical defect, a microvascular fibula free flap from the left side was used, anastomosed to the neck vessels and fixed with a 2.4 mm mandibular reconstruction plate (Figure 7). Uneventful postoperative recovery with adequate mouth opening (Figures 8 and 9). After a 23-year clinical and radiological follow- up, it was possible to observe bone remodelling of the fibula free flap, even at the condylar area, with no evidence of recurrence. Intraorally, mouth opening was preserved, upper and lower dental midlines were aligned and dental attrition due to bruxism was detected.

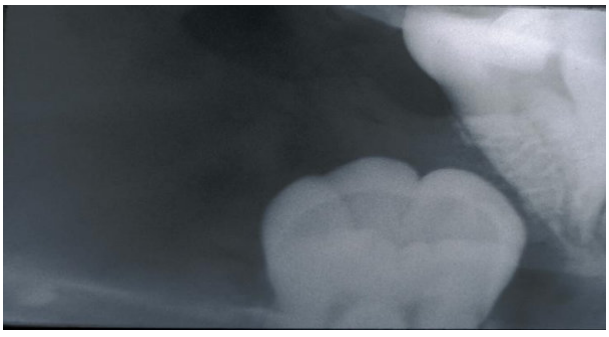

Figure 1. Periapical radiograph showing tooth #38 displaced towards the mandibular basal bone due to an osteolytic radiolucent lesion.

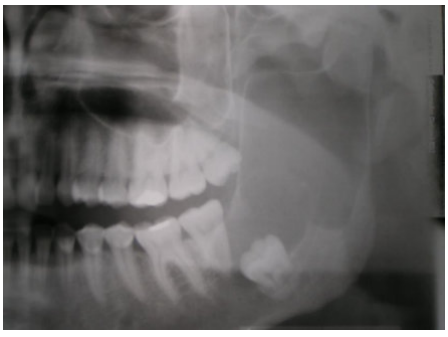

Figure 2. First panoramic radiograph revealing an extensive, unilocular, osteolytic lesion that involves the mandibular ramus, expands the coronoid process and extends up to the condylar neck.

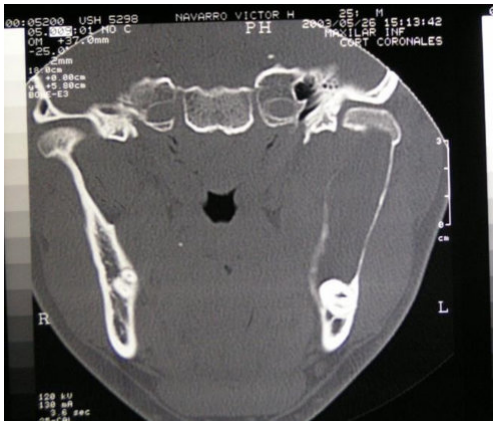

Figure 3. Coronal computed tomography showing severe expansion and thinning of the bone mandibular cortices.

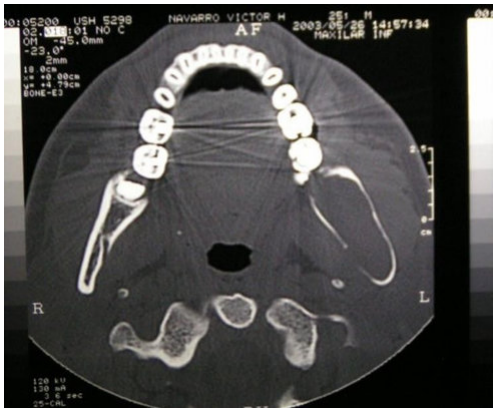

Figure 4. Axial computed tomography showing erosion of the lingual cortex due to the expansive growth of the lesion.

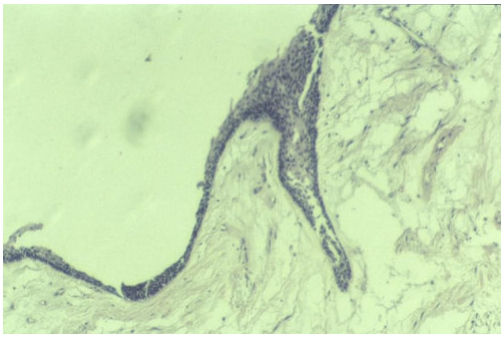

Figure 5. Photomicrograph (H-E 100X) showing epithelial lining of the cystic lesion with intramural growth areas.

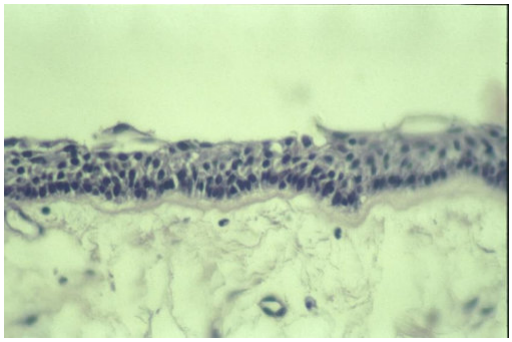

Figure 6. Photomicrograph (H-E 400X) showing Vickers & Gorlin criteria: hyperchromatic basal cells with reverse polarity and palisaded arrangement, intermediate cells with intercellular oedema and flat superficial cells.

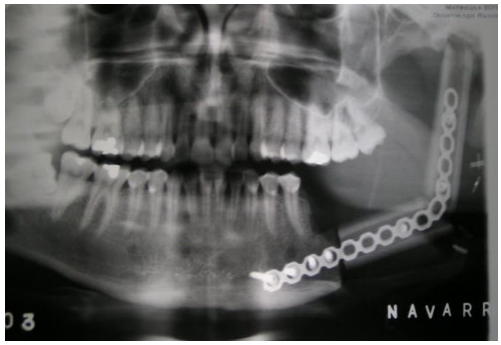

Figure 7. Immediate post-surgical panoramic radiograph where it is possible to appreciate the fibula bone graft adapted to the mandibular body, angle and condyle, fixed using a 2.4 mm mandibular reconstruction plate.

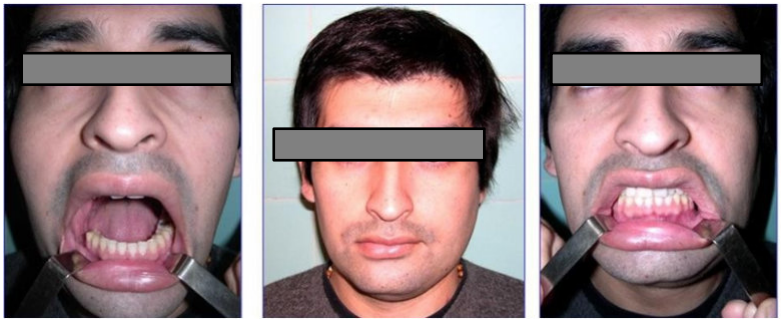

Figure 8. Post-surgical clinical photograph showing an adequate mouth opening range and preservation of mandibular function.

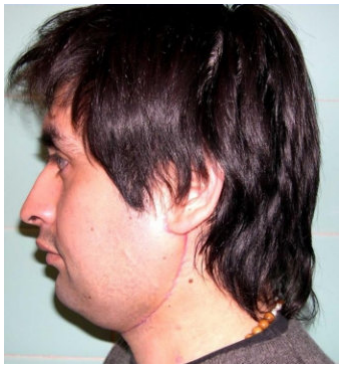

Figure 9. Clinical appearance of the scars after submandibular and preauricular surgical interventions, during healing process.

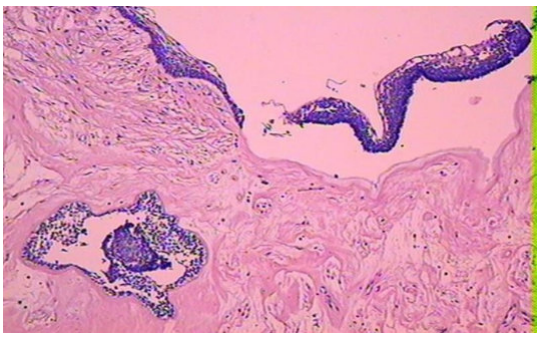

Figure 10. Photomicrograph (H-E 100X) of the resection specimen. Presence of ameloblastic mural nodules infiltrating the cyst wall. H-E 100X.

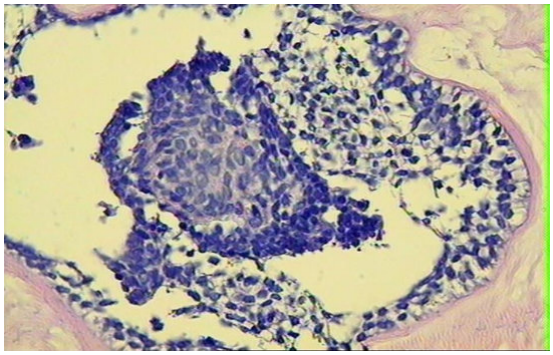

Figure 11. Mural part of an ameloblastoma exhibiting basal cells with reverse polarity, and stellate reticulum-like cells with central squamous metaplasia. HE1,000X.

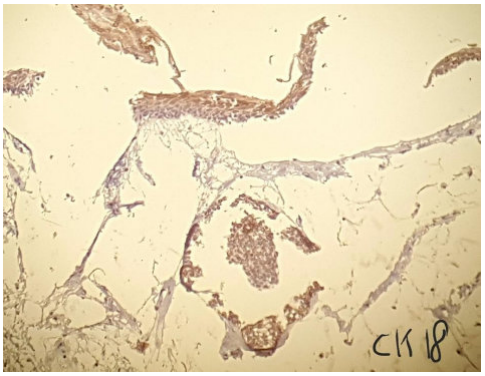

Figure 12. Immunohistochemical study showing strong cytoplasmatic positivity for CK18 in the luminal and mural tumour epithelium. CK18 100X.

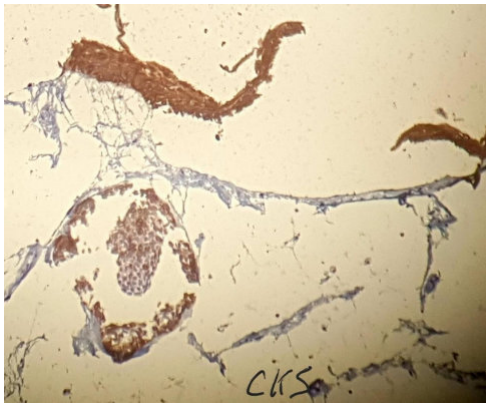

Figure 13. Positive immunohistochemical staining for K5 confirming the odontogenic epithelial lineage of the neoplasm. CK5 100X.

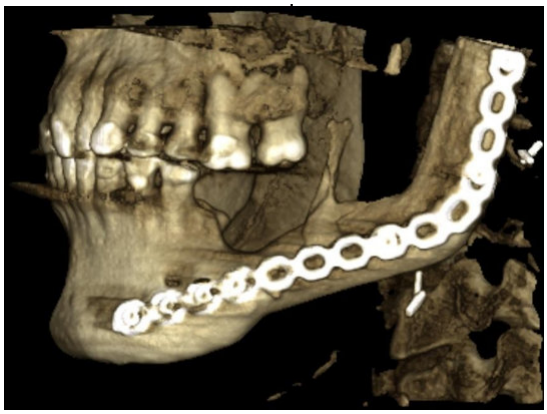

Figure 14. Cone-beam computed tomography at 23 years´ follow-up showing complete osseointegration of the graft and stability of the 2.4 mm reconstruction plate.

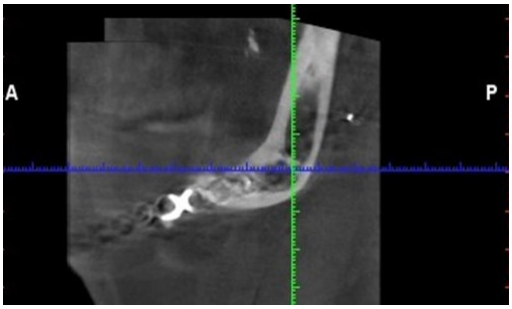

Figure 15. Detailed view of the mandibular angle after 23 years, showing significant bone remodelling of the fibular graft.

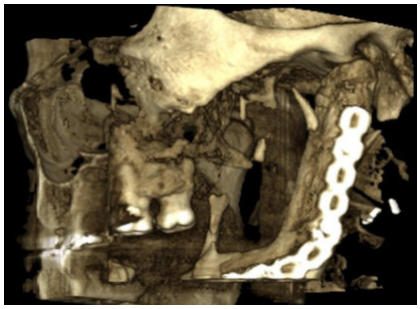

Figure 16. 3D reconstruction using cone-beam computed tomography that illustrates anatomical adaptation of the reconstructed ascending ramus within the glenoid cavity.

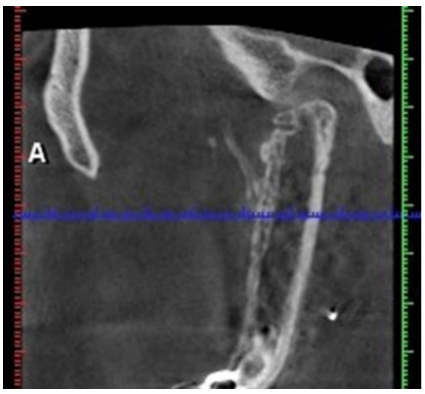

Figure 17. Radiographic detail of the reconstructed left condylar area, showing the blunt morphology acquired by the fibula and resembling a functional condyle.

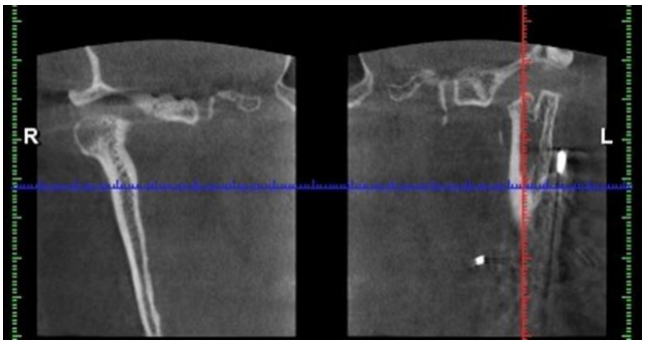

Figure 18. Tomographic comparison showing the symmetry between the native right condyle and the remodelled fibula on the left side.

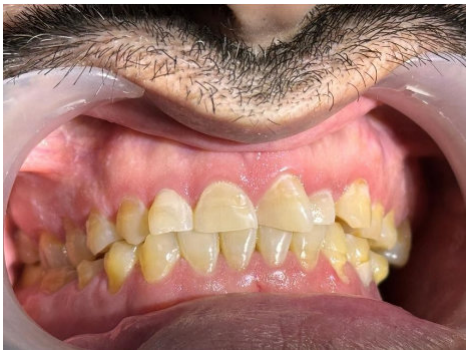

Figure 19. Dental occlusion condition after a two-decade follow-up, which remains functional and stable.

Figure 20. Photograph of maximum mouth opening after a 23-year follow-up, showing signs of dental attrition due to bruxism without affecting the stability of the reconstruction.

Figure 21. Final assessment of the patient´s facial symmetry and mandibular dynamic of the patient after a 23-year follow-up

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Discussion

Unicystic ameloblastoma (UA) was described as one of the subtypes of ameloblastoma in 1977 by Robinson and Martinez. It is a single-cyst neoplasm accounting for between 10% to 15% of all intraosseous ameloblastomas (4,5).

Most UA are asymptomatic. Usually, the first clinical feature is local swelling and pain, especially in the mandibular region of the third molars (6), which is why it may therefore be mistaken for pericoronitis when associated with an impacted tooth, as our patient's case. Another frequent finding is incidental diagnosis via routine radiographic examination, in which a UA may be confused with a dentigerous cyst, keratocyst, periodontal cyst or eruption cyst. Lower lip paraesthesia is rare due to the slow growth rate of the lesion, which produces gradual displacement of the inferior alveolar canal without clinical compromise in most cases, as was the case of the described patient (3).

UA is predominant among young people. There are reports of 7-year-old patients, our 25-year-old patient being consistent with the peak incidence described for this variant. However, exceptional cases have also been reported in patients as old as 70. Depending on the regions, detection is more frequent in young people in the Western countries, while the conventional form of ameloblastoma is more frequent in the African continent (1,2). There is no sex predilection, and more than 50% of the cases are associated with an impacted tooth. Nevertheless, intraosseous location may vary and occur in an interradicular form, resembling periodontal cysts (8), or at the periapical level, resembling radicular cysts or eruption cysts with ameloblastic changes in their wall (9,10). Only one article reported a multilocular lesion, which is controversial given that the word *unicystic* implies a unilocular lesion (9-11). Given the wide variety of intraosseous lesions, a biopsy is mandatory (11,12). Depending on the size of the lesion, the small ones can be totally resected for examination (7). Radiologically, when the lesion is associated with an impacted tooth, it can resemble a dentigerous cyst, especially in the posterior part, where the tooth is displaced with expansion and thinning of the mandibular body and ramus. Currently, studies have been presented regarding its location in the maxilla, the anterior region and invasion into the maxillary sinus (5,13,14).

To confirm UA diagnosis, macroscopically it must present a single sac, which may or may not be related with an impacted tooth. In our case, impacted tooth #38 presented a firm and leathery periodontal ligament surrounding the cementoenamel junction. The epithelium of the cystic lesion must comply with the histological criteria described by Vickers and Gorlin, such as hyperchromatic nuclei, nuclear palisading with reverse polarity, cytoplasmatic vacuolisation resembling ameloblasts, intercellular spacing and oedema in the superficial strata that resembles the stellate reticulum of the tooth germ, flat superficial cells and all these cellular layers resting upon a hyalinisation zone (12). However, the histological subtype is significant to determine a long-term follow-up. Ackerman et al. identified the luminal subtype, in which the histological change is confined to the epithelium of the lesion and presents a low recurrence rate, close to 10%; the intraluminal subtype, in which there is tumour projection into the centre of the cystic cavity and which shares the same recurrence rate as the luminal variant; and the mural subtype, in which tumour growth invades the cystic wall and recurrence rate reaches between 50% and 80% (3). Philipsen and Reichart presented another UA classification: subgroup 1, luminal; subgroup 1-2, luminal and intraluminal; subgroup 1-2-3, luminal, intraluminal and intramural; and subgroup 1-3, luminal and intramural, which is the most widely used classification in the literature (3). Histopathological examination of the case showed a unicystic ameloblastoma with luminal-intramural involvement (subgroup 1-3), both in the biopsy and in the surgical specimen (figures 10-11). Treatment, weather conservative or radical, largely depends on the size of the lesion and the involvement of anatomical structures (15). Enucleation, with or without Carnoy´s solution, is used for small lesions; depending on the final histology of the surgical specimen, follow-up or extension of margins, if needed, is determined, since histological subgroups 1–3 exhibit the highest rate of local recurrence (15). Another variant recently documented in the maxilla is adenoid ameloblastoma, which shows an aggressive behaviour, with a plexiform growth pattern, duct-like structures, epithelial whorls, ghost cell formation and, occasionally, dentinoid deposition (16). The use of immunohistochemistry in biopsies has increased to determine growth factors, aggressiveness, and genetic alterations, such as the presence of BRAF V600E mutation. Its positivity in the specimen allows to consider immunotherapy as part of the treatment and to achieve a reduction in the size of the lesion (17,18).

Immunohistochemistry for CK5 and CK18 was performed, both yielding positive results thus confirming the epithelial odontogenic origin of the lesion (Figures 12-13).

Marginal or segmental resection of the lesion, when adequate bone margins are removed, presents one of the lowest recurrence rates, close to 3.6%. This procedure can be complemented by reconstruction using a bone graft, a reconstruction plate or vascularised free bone grafts such as the fibula or iliac crest (19).

In our case, due to the size of the lesion and its involvement up to the condylar neck, a segmental mandibular resection was chosen, establishing the anterior margin mesial to tooth #36 and including the condyle. For reconstruction, a microvascular fibula free flap was used, which was adapted and remodelled in the part intended to consolidate within the glenoid cavity. Following a 23-year clinical and radiological follow-up, taking a panoramic radiograph and a cone-beam computed tomography, no recurrence was observed. Radiologically, remodelling of the mandibular angle and the condylar portion was observed, the latter showed modelling, corticalisation and adaptation to the glenoid cavity shape, which had not been informed in the literature of previously reported cases where a segmental resection had been performed. In our case, intraoral examination showed stability of the upper and lower dental midlines, centric occlusion, adequate mouth opening range, and dental attrition due to bruxism, which the patient had been suffering over the past year (Figures 14,15,16,17,18,19,20,21).

Maximum follow-up period reported in the literature was 5 years, while minimum was 14 months. Considering the slow-growing nature of the tumour, follow-up should be longer, up to 10 years, and it should also not be limited solely to a panoramic radiograph; it is recommended to also include a tomographic follow-up.

Conclusion

Unicystic ameloblastoma management must be protocolised and personalised according to its extent and histological subtype: enucleation or total resection are recommended for small and confined lesions; while for medium and large ones, prior biopsy is mandatory to determine a specific treatment.

Microvascular fibula free flap is the standard of reference for reconstruction of extensive lesions, offering progressive bone remodelling and long-term functional results, such as the reported 23-year follow-up has shown.

Finally, immunohistochemical marker BRAF V600E must be included in ameloblastoma biopsies protocol to determine the possibility of targeted immunotherapy treatment, which represents the future of biological management of these neoplasms**.**

Conflict of Interest

The authors declare no conflict of interest.

Funding

None

References

1. Ghai S, et al. Ameloblastoma: An updated narrative review of an enigmatic tumor. Cureus. 2022 Aug 6;14(8):e27734.

2. Singh S, et al. Unicystic ameloblastoma: systematic review of clinico-pathologic features and outcomes in infants, children and young adults. Natl J Maxillofac Surg. 2025;16:427-39.

3. Rayamajhi S, Shrestha S, Shakya S, et al. Unicystic ameloblastoma of mandible: a case report. J Nepal Med Assoc. 2022;60(251):657-60.

4. Anchlia S, Bahl S, Vyas S, et al. Unicystic plexiform ameloblastoma with mural proliferation: a full-blown lesion. BMJ Case Rep. 2016.

5. Imase R, Watanabe T. Maxillary unicystic ameloblastoma: a rare presentation. BMJ Case Rep. 2022;15.

6. Karthika P, Kavitha B, Elavenil P. Mural unicystic ameloblastoma mimicking odontogenic cyst. J Family Med Prim Care. 2020 May 31;9(5):2524-7.

7. Ouertani H, et al. Unicystic ameloblastoma: diagnostic challenges and uncommon treatment strategy. Int J Surg Case Rep. 2024;124:110437.

8. Fedhila M, Belkacem C, Karray S, et al. Unicystic ameloblastoma mimicking lateral periodontal cyst. Case Rep Dent. 2022 Feb 14;2022:8197837.

9. Ali M, et al. Unicystic ameloblastoma appearing as massive multilocular entity: a case report with long-term follow-up. Int J Surg Case Rep. 2024;120:109830.

10. McLean A, Vargas PA. Cystic lesions of the jaws: the top 10 differential diagnoses to ponder. Head Neck Pathol. 2023;17:85-98.

11. Bihani N, Byakodi S. Unicystic ameloblastoma: a diagnostic conundrum. Cureus. 2025 Jul 13;17(7): e87870.

12. Vickers R, Gorlin R. Ameloblastoma: delineation of early histopathologic features of neoplasia. Cancer. 1970 Sep;26:699-710.

13. Rajasekaran AM, Prasad C, Balaji J. Unicystic ameloblastoma: a case series. Indian J Dent Res. 2023;34:104-7.

14. Agani Z, Hamiti V, Rexhepi J, et al. Maxillary unicystic ameloblastoma: a case report. BMC Res Notes. 2016;9:469.

15. Kalmegh P, Hande A, Gawande M, et al. Unicystic ameloblastoma (UA): a case series. Cureus. 2022 Nov;14(11):e31039.

16. Martin Chaves R, Cavalieri Gomez C, Fonseca F. Unicystic adenoid ameloblastoma: a new variant? Oral Surg Oral Med Oral Pathol Oral Radiol. 2022;134:e23-28.

17. Joby P, Tharian B, Bijimole J. Unicystic ameloblastoma associated with primary mandibular second molar: a case report. Int J Clin Pediatr Dent. 2023;16(Spec Issue 3).

18. Pattnaik B, Mohanty S, Das S, et al. Immunohistochemical evaluation of yes-associated protein molecule in the odontogenic epithelium of different histopathological variants of ameloblastoma and unicystic ameloblastoma. J Oral Maxillofac Pathol. 2024;28:49-55.

19. Kalwagadda S, Kumar B, Nair S, et al. Management of ameloblastoma with free tissue flap in comparison with other reconstructive options available. J Maxillofacial Oral Surg. 2020;19:283-8.