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CASE REPORT

MANDIBULAR RECONSTRUCTION WITH ILIAC CREST GRAFT FOR RECURRENT AMELOBLASTOMA: A CASE REPORT

Reconstrucción mandibular con injerto de cresta ilíaca en ameloblastoma recidivante: informe de un caso

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ABSTRACT

Background: Ameloblastoma is an odontogenic tumor characterized by slow but aggressive growth, which can lead to significant facial deformity and functional impairment.

Case Report: We present the case of a 47-year-old patient with a history of mandibular ameloblastoma treated with enucleation and curettage 20 years ago. The patient presented pain, paresthesia of the lower lip, and swelling in the mandibular region. Panoramic radiography and cone-beam computed tomography revealed a multilocular osteolytic lesion in the left hemimandible, suggesting tumor recurrence. An incisional biopsy confirmed the diagnosis of conventional ameloblastoma, plexiform variant. Due to the recurrent nature of the tumor, a radical approach was taken, consisting of block resection of the affected hemimandible followed by mandibular reconstruction using an autologous iliac crest graft. The surgery was meticulously planned using stereolithographic models, allowing precise adaptation of the graft. Postoperative follow-up showed positive results with complete healing and adequate functional recovery.

Conclusions: An accurate diagnosis is essential to determine the appropriate treatment for ameloblastoma, and thorough planning ensures a successful surgical outcome. The use of a free iliac crest graft offers several advantages, such as low morbidity at the donor site, availability of corticocancellous bone, and the ability to restore both function and aesthetics. Favorable results were achieved, significantly improving the patient's quality of life.

Keywords: *Ameloblastoma; Autologous transplant; Iliac crest; Mandibular reconstruction; Case report*

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RESUMEN

Introducción: El ameloblastoma es un tumor odontogénico caracterizado por un crecimiento lento pero agresivo que puede provocar deformidades faciales significativas y problemas funcionales.

Reporte de Caso: Se presenta el caso de una paciente femenina de 47 años con antecedentes de ameloblastoma mandibular tratado hace 20 años mediante enucleación y curetaje. La paciente acudió por dolor, parestesia del labio inferior y aumento de volumen en la región mandibular. Una radiografía panorámica y una tomografía cone beam revelaron una lesión osteolítica multilocular en la hemimandíbula izquierda, sugerente de recidiva tumoral. Se realizó una biopsia incisional, que confirmó el diagnóstico de ameloblastoma convencional variante plexiforme. Dada la naturaleza recidivante del tumor, se optó por un tratamiento radical que consistió en una resección en bloque de la hemimandíbula afectada, seguido de reconstrucción mandibular con injerto autólogo de cresta ilíaca. La cirugía fue planificada utilizando modelos estereolitográficos, lo que permitió una adaptación precisa del injerto. El seguimiento postoperatorio mostró resultados positivos , con cicatrización completa y recuperación funcional adecuada.

Conclusión: El diagnóstico correcto es fundamental para establecer el tratamiento del ameloblastoma, una minuciosa planificación permite una ejecución adecuada del procedimiento quirúrgico, el injerto libre de cresta iliaca presenta ventajas como baja morbilidad del sitio donante, disponibilidad de hueso corticoesponjoso, y recuperar la función y estética. Se obtuvo resultados favorables devolviendo la calidad de vida a la paciente.

Palabras clave: *Ameloblastoma; Trasplante autólogo; Cresta iliaca; Reconstrucción mandibular; Informe de caso*

INTRODUCTION

Ameloblastoma is the most common odontogenic tumor, characterized by slow but aggressive growth that can potentially lead to significant facial deformities and functional impairments if not adequately treated.[1,2]

This benign but locally aggressive neoplasm originates from the ectomesenchyme, derived from remnants of the dental lamina, enamel organ, or the lining of an odontogenic cyst.[2,3] Clinically, it typically presents as a painless swelling in the mandible, most often in the posterior region, and can be associated with cortical perforation, displacement of dental structures, paresthesia, and is frequently linked to impacted teeth.[2,4]

The World Health Organization 2022 classification of odontogenic cysts and tumors recognizes five distinct types of ameloblastoma: unicystic, peripheral, conventional, adenomatoid and metastasizing.[5] Conventional ameloblastoma exhibits various histological patterns based on McMilan's 1971 classification, with the follicular type being the most common (24.8%), followed by plexiform (24.7%), acanthomatous (5.7%), granular cell (2.5%), and basal cell (0.4%).[4] Among these, the plexiform type is the second most common, with a recurrence rate of 15.1%.[6]

Radiographically, ameloblastoma presents with variable characteristics, ranging from a unilocular radiolucent shadow to multilocular patterns resembling a honeycomb appearance with well-defined corticated margins. Advanced imaging studies such as computed tomography can demonstrate cortical expansion and reveal details of the internal bone architecture.[1,4] The global incidence of ameloblastoma is estimated at 0.5 cases per million people per year.[1]

However, its prevalence varies significantly between geographical regions, being significantly higher in Africa and Asia, where it represents up to 43% of maxillomandibular tumors.[7] In North America, the reported incidence is 0.21%.[8] In Latin America, comprehensive frequency studies remain scarce, but available data suggests a range between 5.8% and 34.6%, with predominant involvement of individuals in their third decade of life and no significant differences between sexes.[1,7] Epidemiological studies indicate that Afro-Americans demonstrate a five-fold higher likelihood of developing this pathology compared to Caucasians.[7]

Treatment approaches for ameloblastoma can be broadly categorized as conservative or radical.[9] Conservative treatment options include marsupialization, enucleation, curettage, enucleation combined with Carnoy's solution application, enucleation combined with curettage, and curettage combined with cryotherapy.[10,11] However, these conservative approaches are associated with high recurrence rates of up to 40%, primarily attributed to tumor cell infiltration beyond the radiographically visible boundary.[9] In contrast, radical treatment approaches, including segmental or marginal resections of the mandible and partial or total maxillectomies, require adequate bone margins of 1 to 1.5 cm to effectively minimize the risk of recurrence.[2,10]

Mandibular reconstruction represents a crucial component of comprehensive treatment after radical resection of ameloblastoma, with the primary goal of restoring both functional capacity and facial aesthetics.[6] Among these options, the iliac crest is considered a suitable graft source that reduces the complexity associated with multiple osteotomies and vascular anastomoses compared to fibula grafts.[12,13] Additionally, in cases requiring vertical height augmentation, distraction osteogenesis represents another viable reconstructive option.[6,10]

The management of recurrent ameloblastoma poses particular challenges due to the aggressive nature of these tumors and the complexity of revision surgery. This clinical case underscores the importance of radical management in the treatment of recurrent aggressive tumors such as ameloblastoma, highlighting the value of meticulous surgical planning that incorporates advanced techniques such as stereolithographic modeling to ensure precise graft adaptation and optimize surgical outcomes. Furthermore, this report aims to contribute valuable information on the surgical outcomes of this reconstructive approach to the wider scientific community. The purpose of this article is to describe the surgical management of recurrent ameloblastoma through block resection and reconstruction with an iliac crest graft.

CASE REPORT

A 47-year-old female patient came to the Maxillofacial Surgery department with a major complaint of pain, paresthesia of the lower lip, and progressive swelling in the left mandibular region. The patient had a history of a mandibular tumor treated 20 years ago with enucleation and curettage. At the time of the current consultation, a panoramic radiograph revealed a multilocular radiolucent shadow with a honeycomb-like appearance, involving the body and ramus of the left mandible (Figure 1A).

Physical examination revealed swelling in the left hemimandible, accompanied by a fistula in the oral mucosa, suggesting a possible tumor recurrence (Figure 2). A cone beam computed tomography scan showed a hypodense multilocular image with hyperdense halos, consistent with a 5.3 x 4 cm osteolytic lesion, along with cortical expansion and vestibular cortical lysis (Figure 1B and Figure 1C). Based on these findings, an incisional biopsy of the lesion was performed under local anesthesia, along with mucosal plasty.

Histopathological analysis confirmed

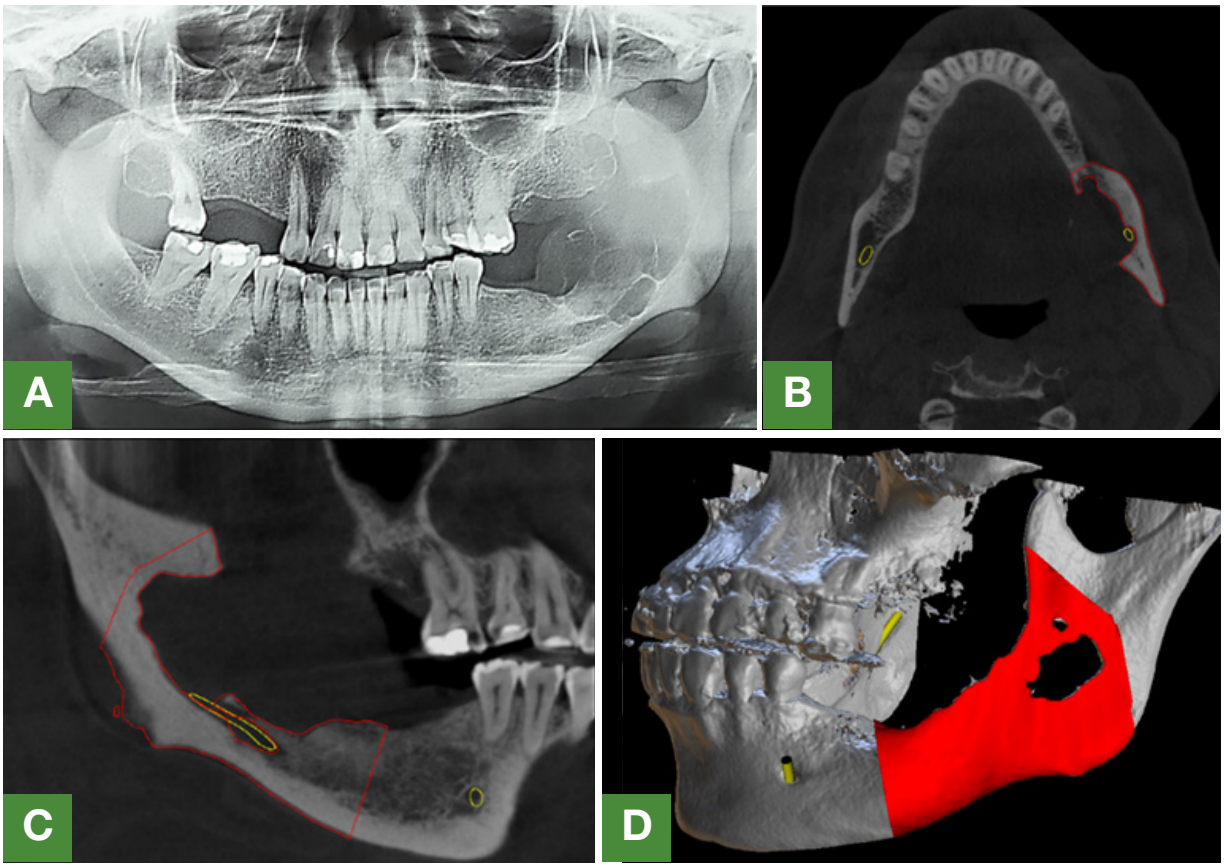
the diagnosis of conventional ameloblastoma, plexiform variant.

Given the recurrent nature of the tumor, a radical surgical intervention was planned, which included segmental resection of the left hemimandible, involving both the body and the ramus with safety margins (Figure 1D), along with reconstruction using an iliac crest graft. Before surgery, stereolithographic models of the mandible were created and a custom reconstruction plate was prepared (Figure 3).

The surgical procedure was performed under general anesthesia with nasotracheal intubation, using a Risdon-type extraoral approach. The lesion was defined with 1.5 cm safety margins and the affected mandibular segment was resected in block using a reciprocating saw. Then, a 2.4mm pre-constructed reconstruction plate system (LeFórte, South Korea) was placed. During the same time, the trauma team harvested a 6 cm iliac crest graft, which was adapted to the mandibular bone defect and secured with osteosynthesis screws (Figure 4).

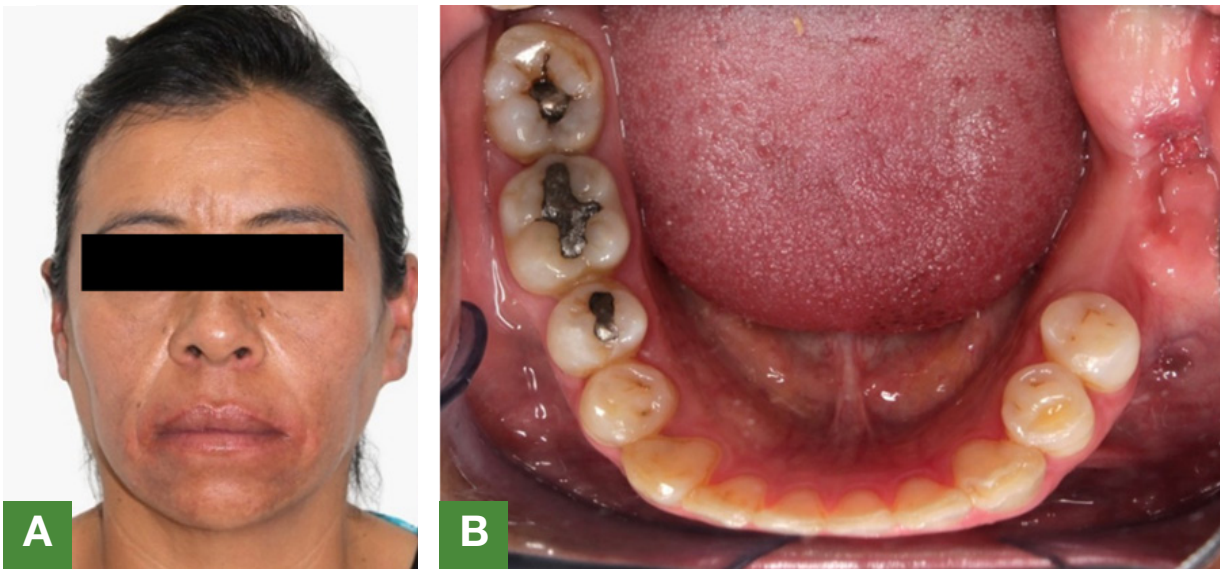
Finally, the surgical approach was closed in layers, using 4-0 polyglactin sutures for the deep layers and 5-0 polypropylene sutures for the superficial layer. In the immediate postoperative period, the patient remained hospitalized for five days for pain management and antibiotic therapy (ampicillin-sulbactam 3 g intravenously every 6 hours for 5 days, ketorolac 30 mg intravenously every 8 hours for 4 days, paracetamol 1 g intravenously every 8 hours for 5 days, dexamethasone 8 mg intravenously for 3 days), as well as daily dressing changes of the surgical wound. The

Figure 1
A cone Beam Computed Tomography



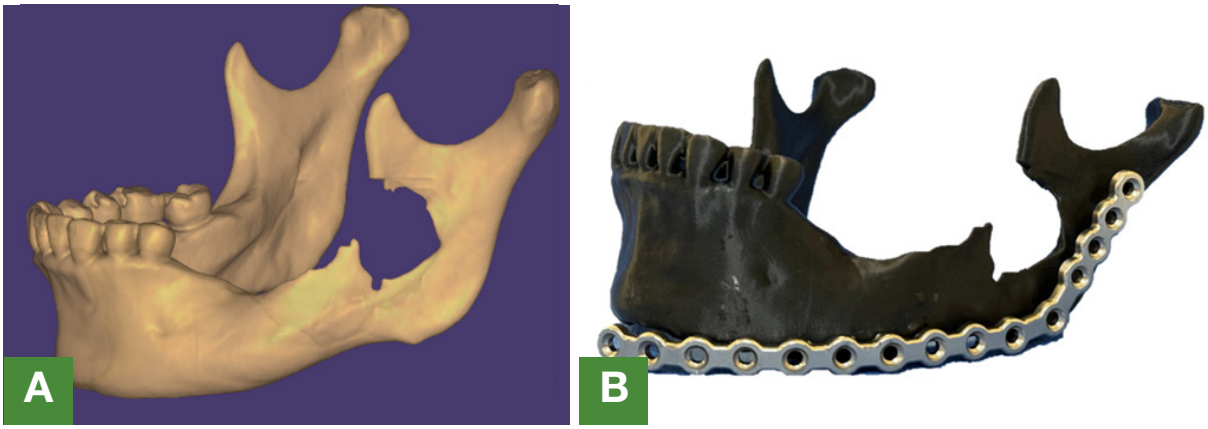
A: Panoramic radiograph. **B:** Axial slice computed tomography of cone beam. **C:** Sagittal slice of cone beam computed tomography. **D:** 3D reconstruction delimiting the margins of the lesion.

Figure 2
Physical examination suggesting a possible tumor



A: Extraoral photograph. **B:** Intraoral photograph showing a fistula towards the lesion.

Figure 3
Stereolithographic models of the mandible before surgery and reconstruction plate



A: 3D reconstruction. **B:** Stereolithographic model with preformed plate.

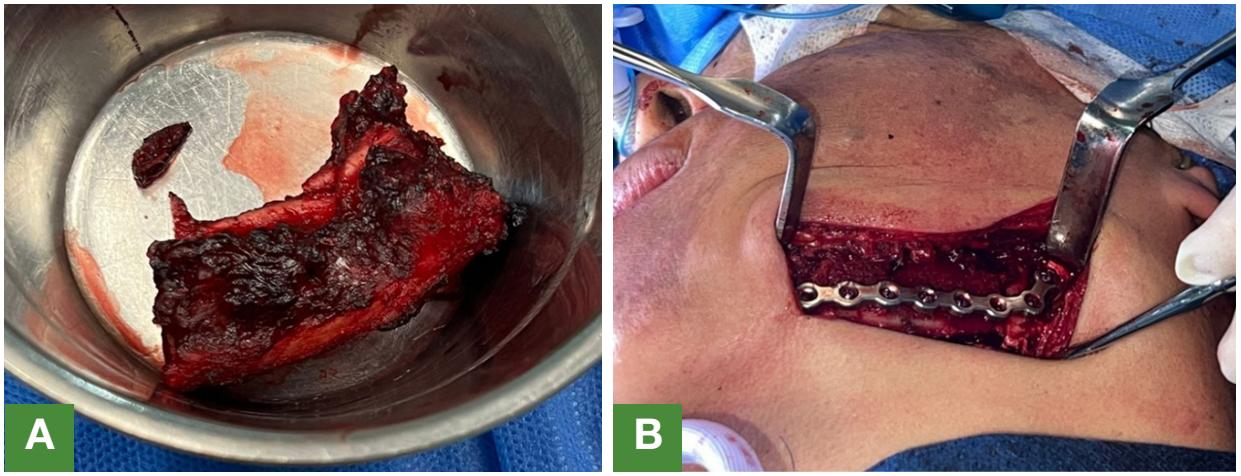
recovery of the patient was favorable, with mild edema and adequate pain control.

At eight days of postoperative follow-up, the surgical wound appeared clean with epithelialization in progress and decreasing edema. After 15 days, the patient showed a favorable evolution with stable dental occlusion and complete wound healing, allowing suture removal. At 30 days, a panoramic radiograph demonstrated the proper position of the bone graft and the osteosynthesis material (Figure 5A and Figure 5B). At four months, complete wound healing was reported and the patient reported satisfactory masticatory function. Following the completion of a one-year follow-up, observations indicate excellent graft integration, stable dental occlusion, and satisfactory facial aesthetics (Figure 5C and Figure 5D).

The patient reports satisfactory masticatory function devoid of pain or functional limitations. Nonetheless, mild paresthesia

Figure 4

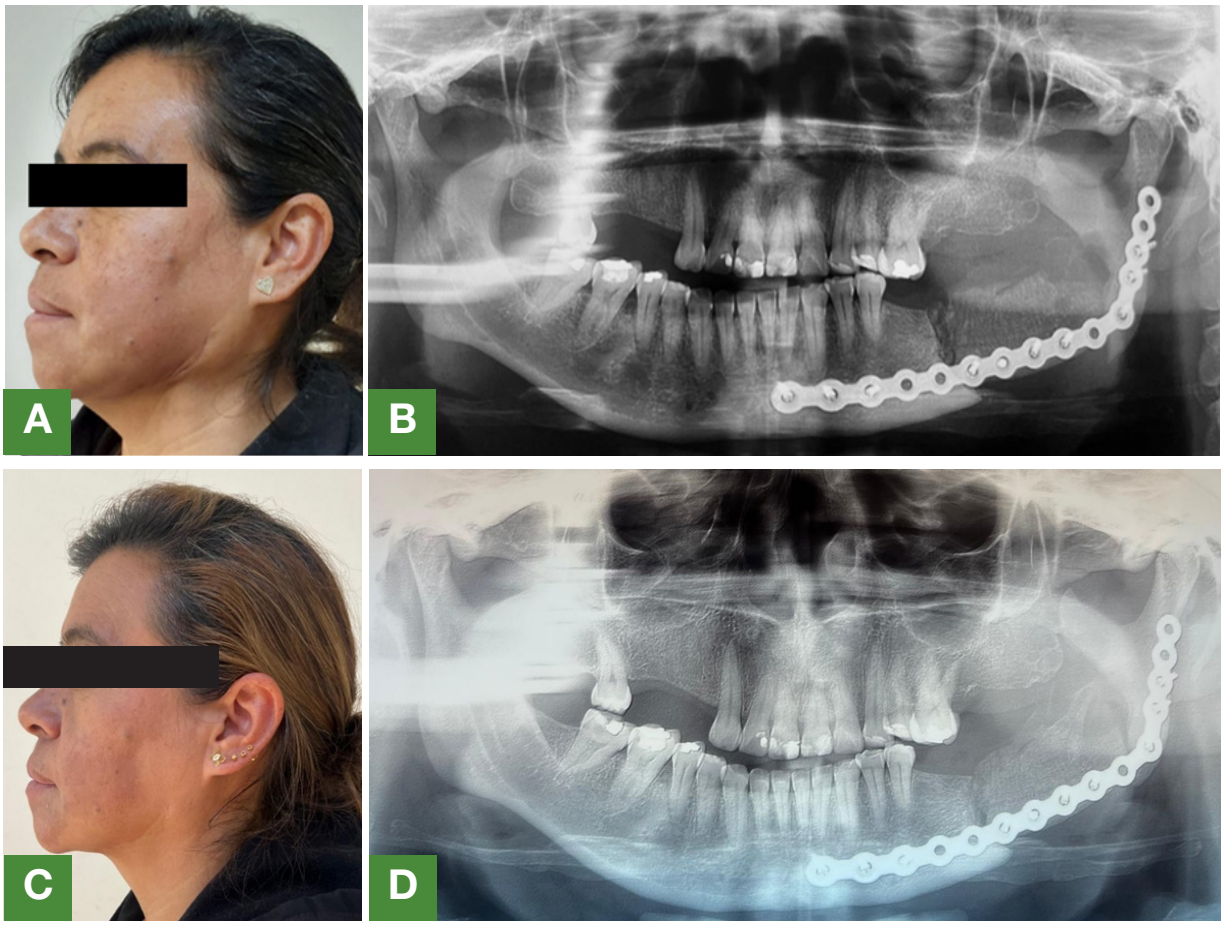
The surgical procedure and reconstruction plate system pre-constructed



A: Tricortical iliac crest graf. **B:** The graft was adapted to the recipient site in the mandible, correctly positioned, and fixed with osteosynthesis material.

Figure 5

Postoperative follow-up



A: 1-month extraoral follow-up photograph. **B:** 1-month follow-up panoramic radiograph shows correct integration of the graft. **C:** 12-month extraoral follow-up photograph. **D:** 12-month follow-up panoramic radiograph shows correct integration of the graft.

of the left lower lip and chin persists, yet it does not significantly impact the patient's quality of life.

The initiation of dental implant rehabilitation is contingent upon the full osseo-integration of the graft. The advised os-seointegration period, which spans 6 to 12 months, has been fulfilled, and the subsequent implant placement is projected for the forthcoming months. Nonetheless, the scheduling of treatment is subject to financial constraints, given that the surgical procedure was conducted within a public healthcare framework, where dental implants are excluded from the standard service offerings.

ETHICAL CONSIDERATIONS

The present case report was prepared after the patient signed the informed consent form and agreed to the treatment plan. Furthermore, the patient approved the use of clinical information and photographs to be anonymously presented to a scientific journal. The case has been approved by the research committee of the Faculty of Dentistry of the Central University of Ecuador.

DISCUSSION

The surgical management of recurrent plexiform ameloblastoma presents a significant clinical challenge due to the aggressive nature of this tumor and its high recurrence potential.[14] In this case, given the large size of the lesion (5.3 x 4 cm) and its recurrent nature after 20 years, a radical treatment approach was chosen, which involved segmental resection of the affected hemimandible and immediate reconstruction with an iliac crest graft. This approach allowed adequate tumor excision with sufficient safety margins and reconstructive surgery that restored both function and aesthetics for the patient.

Treatment of ameloblastoma varies depending on its clinical and histological presentation. A conservative approach is recommended for unicystic and peripheral tumors, which are typically treated with techniques such as curettage, enucleation, and cryosurgery, due to their lower recurrence rate, reported to be 3% in some studies. In contrast, solid or multilocular ameloblastomas, which are considered more aggressive, require radical treatment, including reconstructive surgery, due to their higher recurrence potential, which can reach up to 41% with conservative treatment. Several studies have confirmed that follicular, plexiform, and mixed tumors have higher recurrence rates (25-33%), highlighting the importance of considering histological type in therapeutic decision-making.[9-11]

Segmental resection achieved the highest score in reducing recurrence rates (77.7%), followed by curettage combined with cryotherapy and marginal resection. Cryotherapy induces cellular necrosis through freezing, enabling the eradication of residual tumor cells without compromising the integrity of the inorganic bone structure.[9,11] Conservative approaches due to higher recurrence rates, leading to the need for reinterventions. In contrast, radical surgical, present lower recurrence rates but often result in aesthetic and functional sequelae that impact the patient's quality of life and require complex reconstructive procedures.[9,11,16]

Previous studies have reported that the rate of ameloblastoma recurrence is slightly higher in women, with frequent presentation in the third and fourth decades of life.[6,17]

These findings are consistent with the case presented here, which involves a 47-year-old patient.[9] In this case, the recurrent plexiform variant justified the need for a radical intervention to minimize the risk of recurrence. Hendra *et al.*,[11] demonstrated that segmen-

tal resection is the most effective option, although the approach should be tailored to the anatomical characteristics and size of the tumor.

The purpose of the graft is to restore masticatory, phonatory and respiratory functions while preserving dental occlusion and temporomandibular joint function, which ultimately allows dental rehabilitation.[18] The choice of graft is based on the length of the defect, the recipient site, the availability of blood vessels if anastomosis is required, and the quality of bone and soft tissue.[12,20] Mandibular defects can be reconstructed with autografts, allografts, or alloplastic grafts. Autografts have superior characteristics due to their lower immunological response, which reduces the risk of rejection and infection. Autografts can be vascular bone grafts or nonvascular bone grafts.[21]

Due to the advancement of surgical techniques, microvascularized free flaps are currently the gold standard for reconstructive surgery, while nonvascular grafts may be an option in patients who do not require radiotherapy and who have a sufficient quantity and quality of remaining bone and soft tissue.[12] The iliac crest represents a suitable graft option due to several advantageous characteristics: length and height similar to the mandible, ample availability of cancellous bone and osteoprogenitor cells, and a blood supply that reduces the likelihood of infection and improves implant osseointegration. The donor site has a less complex anatomy and is easily accessible.[20,21]

However, among its limitations, it presents a high degree of resorption, multiple osteotomies can compromise its vascularity, and it is preferable for defects up to 6 cm. Finally, the donor site is not exempt from complications, such as hematomas, inguinal hernia, gait disturbance, or prolonged pain.[20,21]

The free fibula graft is a suitable option as a vascularized bone graft, it provides 20-25 cm of available bone to reconstruct large defects (greater than 9 cm), allows multiple osteotomies to recontour the mandible without compromising its viability, and allows the placement of dental implants.[18,20,21] Among its disadvantages are a longer operating time due to microanastomosis and recontouring for graft adaptation. Harvesting must be performed by experienced surgeons to minimize donor site morbidity. The patient requires a longer recovery time due to restricted movement and postoperative walking difficulties. The general health of the patient can also be a limiting factor, for example, in cases of circulatory conditions such as varicose veins or peripheral vascular disease.[12,22]

With technological advancements, concepts such as "jaw-in-a-day" have emerged. This process allows patients to receive reconstructive surgery in a single day, including tumor resection, graft placement, dental implants, and prosthetics. These technological improvements were pioneered by Levine et al., who in 2013 created a digital workflow to design dental prosthetics and cut guides for the fibula incorporating holes for dental implant placement.[23] This approach offers numerous advantages for patients, such as fewer surgical interventions, improved soft tissue healing, shorter rehabilitation time, and reduced psychosocial impact. Currently, this type of surgery is more common in the United States; however, it remains limited in developing countries due to high costs and the shortage of experienced surgical teams.[23,24]

Although radical treatment is the approach of choice for conventional ameloblastoma, initial conservative treatment in this case maintained function and aesthetics for an extended period. However, considering the aggressive characteristics of this tumor, radical treatment becomes essential in cases of recurrence, requiring longer follow-up pe-

riods with clinical and radiographic controls, which represents a limitation in this case given our relatively short follow-up period.[11]

In delayed implant rehabilitation of bone grafts of the iliac crest, a waiting period of 6 to 12 months is generally recommended, depending on the degree of osseointegration of the graft and the patient's systemic and local conditions.[12] In the present clinical case, the recommended integration period has been completed, and implant placement is planned in the coming months. However, since the surgical procedure was performed in a public healthcare setting and dental implants are not included in the standard service portfolio, financial constraints must be considered as a determining factor in the timing and feasibility of implant treatment.

One of the strengths of this case was the use of a multidisciplinary approach that combined precise surgical planning with advanced reconstruction techniques, incorporating stereolithographic models that allowed accurate graft adaptation and reduced surgical time, resulting in successful functional and aesthetic recovery. However, a limitation of this report is that the one year follow-up period is relatively short. Although the results are promising, this period is insufficient to definitively assess long-term stability of the graft and the risk of tumor recurrence, as ameloblastoma recurrences typically occur within five years.[11] Another limitation is that the lack of immediate dental implant rehabilitation prevents an evaluation of complete functional restoration.

This case highlights the importance of adopting a radical approach to the management of recurrent ameloblastomas, particularly the conventional plexiform variant. Although the results obtained are promising, this is based on a single case, which limits the generalizability of the findings. More studies, such as prospective studies or clinical trials, are required to evaluate the long-term stability of different grafts and the absence of tumor recurrence. Furthermore, it would be beneficial to explore the application of adjuvant therapies, such as molecular therapy, radiation therapy, or chemotherapy, in the cases of highly recurrent ameloblastomas or those with aggressive histopathological characteristics.

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CONFLICT OF INTERESTS

The authors declare that they have no conflicts of interest related to this scientific article.

ETHICS APPROVAL

Informed consent was obtained from the patient, and the case report has been approved by the research committee of the Faculty of Dentistry of the Central University of Ecuador.

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AUTHORS' CONTRIBUTIONS

Karol Romero: Concept development, study design, definition of intellectual content, literature search, data acquisition, data analysis, manuscript preparation, manuscript editing, manuscript review, and served as guarantor of the work.

Eduardo Ramos: Concept development, study design, definition of intellectual content, literature search, data acquisition, data analysis, manuscript preparation, manuscript editing, manuscript review, and served as guarantor of the work.

Juan Marcos Parise-Vasco: Study design, definition of intellectual content, literature search, manuscript preparation, manuscript editing, and manuscript review.

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