Microvascularized fibula reconstruction for the treatment of recurrent primary meningioma: a case report with seven years of follow up

Reconstrução microvascularizada da fíbula para o tratamento de meningioma primário recorrente: relato de caso com sete anos de acompanhamento

Reconstrucción microvascularizada de la fíbula para el tratamiento del meningioma primario recurrente: relato de caso con siete años de acompañamiento

DOI:10.34119/bjhrv8n5-112

Submitted: Aug 22th, 2025
Approved: Sep 12th, 2025

Robson Rodrigues Garcia
PhD in Oral and Maxillofacial Surgery
Institution: Universidade Federal de Goiás
Address: Goiânia, Goiás, Brazil
E-mail: robsongarcia@ufg.br

Thais Rodrigues da Silveira
PhD in Oral and Maxillofacial Surgery
Institution: Universidade Federal de Goiás
Address: Goiânia, Goiás, Brazil
E-mail: thaissilveira@ufg.br

Gileade Pereira Freitas
PhD in Oral and Maxillofacial Surgery
Institution: Universidade Federal de Goiás
Address: Goiânia, Goiás, Brazil
E-mail: gileade@ufg.br

Roger William Fernandes Moreira
PhD in Oral and Maxillofacial Surgery
Institution: Carolinas Center for Oral and Facial Surgery
Address: Raleigh, United States of America
E-mail: cirurgiarm@gmail.com

Maria Eduarda Cardoso Inácio
Masters Candidate of the Graduate Program Faculdade de Odontologia of Universidade Federal de Goiás (FO-UFG)
Institution: Universidade Federal de Goiás
Address: Goiânia, Goiás, Brazil
E-mail: mariainacio@discente.ufg.br

Laura Olímpio Serrano Mendes
Masters Candidate of the Graduate Program Faculdade de Odontologia of Universidade Federal de Goiás (FO-UFG)
Institution: Universidade Federal de Goiás
Address: Goiânia, Goiás, Brazil
E-mail: lauraolimpio@discente.ufg.br

Gabriella Rodrigues Garcia
DMD, Brazilian Federal Board of Dentistry Certified Orthodontist
Instituição: Orthodontic Private Clinic in Goiânia
Endereço: Goiânia, Goiás, Brazil

ABSTRACT
Meningiomas are typically benign tumors of mesodermal origin and are recognized as the most common extra-axial neoplasms of the central nervous system. Although they predominantly arise within the cranial cavity, a small subset occurs extracranially. Primary extracranial meningiomas located in the maxillomandibular region are extremely rare, often presenting diagnostic challenges due to their resemblance to other soft tissue or bone lesions. The surgical management of primary meningiomas at the mandible may require segmental resection. In such cases, vascularized fibular bone grafts offer an ideal reconstructive solution, when the iliac crest is not enough allowing for adequate bone length, reliable vascular supply, and compatibility with future prosthetic rehabilitation. This approach also supports three-dimensional mandibular contour restoration and soft tissue integration. This article presents a case involving the late recurrence of a previously managed primary extracranial meningioma in the mandible. It outlines the surgical strategy and outcomes, underlining the critical role of long-term monitoring in rare oncologic presentations within the maxillofacial region.

Keywords: microvascularized fibula reconstruction, primary extracranial meningioma, mandibular segmental resection, vascularized bone graft.

RESUMO
Os meningiomas são tumores tipicamente benignos de origem mesodérmica e são reconhecidos como as neoplasias extra-axiais mais comuns do sistema nervoso central. Embora surjam predominantemente na cavidade craniana, um pequeno subconjunto ocorre extracranialmente. Os meningiomas extracranianos primários localizados na região maxilomandibular são extremamente raros, apresentando frequentemente desafios diagnósticos devido à sua semelhança com outras lesões dos tecidos moles ou ósseos. O tratamento cirúrgico dos meningiomas primários na mandíbula pode exigir ressecção segmentar. Nesses casos, os enxertos ósseos fibulares vascularizados oferecem uma solução reconstrutiva ideal, quando a crista ilíaca não é suficiente para permitir comprimento ósseo adequado, suprimento vascular confiável e compatibilidade com futura reabilitação protética. Essa abordagem também favorece a restauração tridimensional do contorno mandibular e a integração dos tecidos moles. Este artigo apresenta um caso envolvendo a recorrência tardia de um meningioma extracraniano primário previamente tratado na mandíbula. Ele descreve a estratégia cirúrgica e os resultados, destacando o papel crítico do monitoramento de longo prazo em apresentações oncológicas raras na região maxilofacial.

Palavras-chave: reconstrução microvascularizada da fíbula, meningioma extracraniano primário, ressecção segmentar da mandíbula, enxerto ósseo vascularizado.

RESUMEN
Los meningiomas son tumores típicamente benignos de origen mesodérmico y se reconocen como las neoplasias extraaxiales más comunes del sistema nervioso central. Si bien se originan predominantemente en la cavidad craneal, un pequeño subgrupo se presenta extracranealmente. Los meningiomas extracraneales primarios ubicados en la región maxilomandibular son extremadamente raros y a menudo presentan dificultades diagnósticas debido a su similitud con otras lesiones óseas o de tejidos blandos. El tratamiento quirúrgico de los meningiomas primarios mandibulares puede requerir una resección segmentaria. En estos casos, los injertos óseos vascularizados de peroné ofrecen una solución reconstructiva ideal cuando la cresta ilíaca no es suficiente para lograr una longitud ósea adecuada, un aporte vascular fiable y la compatibilidad con futuras rehabilitaciones protésicas. Este abordaje también facilita la restauración tridimensional del contorno mandibular y la integración de los tejidos blandos. Este artículo presenta un caso de recurrencia tardía de un meningioma extracraneal primario mandibular previamente tratado. Se describe la estrategia quirúrgica y los resultados, destacando la importancia de la monitorización a largo plazo en casos oncológicos poco frecuentes en la región maxilofacial.

Palabras clave: reconstrucción microvascularizada del peroné, meningioma extracraneal primario, resección segmentaria mandibular, injerto óseo vascularizado.

1 INTRODUCTION

Meningiomas are benign neoplasms of mesodermal origin and constitute the most common extra-axial tumors of the central nervous system (CNS) (Mosqueda-Taylor, 2009). These lesions arise from arachnoid cells that infiltrate the dura mater to form arachnoid granulations, typically located near the entry points of blood vessels and nerves (Mosqueda-Taylor, 2009). While the majority of cases are intracranial, extracranial meningiomas are rare, accounting for approximately 2% of all occurrences (Rege, 2017).

Primary extracranial meningiomas involving the maxillomandibular region are extremely rare, which can complicate differential diagnosis with other lesions (Rege, 2017; Iaconetta, 2012). When present in this area, they tend to affect women, with an average age of 35.9 years, typically presenting as a painless swelling on the right side of the mandible (Dionísio, 2020). However, due to the limited number of cases reported in the literature, definitive demographic conclusions cannot yet be established. Moreover, long-term follow-up and rehabilitation treatment reports are scarce, which restricts understanding of the biological behavior, potential complications, and recurrence risk associated with this manifestation of the disease (Dionísio, 2020).

Extended segmental resection is indicated in cases of extensive mandibular bone involvement, as seen in the reported tumor (Ooi, 2014; Chen, 2019; Mertens, 2014; Archual, 2022). Mandibular reconstruction using a vascularized fibular bone graft is considered the optimal treatment following segmental resections, due to its favorable bone length, reliable vascular supply, and compatibility with prosthetic rehabilitation (Tarsitano, 2023; Chen, 2019; Hidalgo, 2002). Notably, this approach enables three-dimensional mandibular reconstruction, providing adequate support for soft tissues and offering strong potential for implant-based rehabilitation (Manju, 2023; Tarsitano, 2023).

This article aims to report the late recurrence of a previously treated primary extracranial meningioma, as well as to describe the therapeutic approach adopted and the postoperative outcomes, highlighting the importance of longitudinal follow-up in rare oncological cases of the maxillofacial region.

2 CASE REPORT

A 48-year-old male patient was diagnosed in August 2011 with a primary extracranial meningioma affecting the body and ramus of the right mandible. Diagnostic details are documented in the case report “Primary Extracranial Meningioma: A Rare Location” by Rege, Inara Carneiro Costa *et al*. (Inara, 2017), published in the *Journal of Head and Neck Pathology*. At the time, imaging revealed that the lesion involved both the mandibular body and ramus on the right side (Fig. 1). An incisional biopsy was performed, and the diagnosis was confirmed through histopathological and immunohistochemical analysis, which showed positive staining for EMA, vimentin, and CD34 antibodies, with no reactivity for other markers.

In the following month, the patient underwent segmental resection of the mandible. The excised surgical specimen measured approximately 5 cm at its greatest diameter and included the mandibular ramus, angle, the inferior alveolar nerve, and the second lower molar. During the same procedure, reconstruction was performed using an autogenous tricortical iliac bone graft, secured with a 2.4 mm locking plate system (Fig. 2).

2.1 RECURRENCE AND NEW SURGICAL APPROACH

The patient was monitored weekly during the first month, followed by monthly follow-up visits until the sixth month. At the one-year mark, he returned without any significant signs or symptoms and underwent a computed tomography (CT) scan, which confirmed the success

of the surgical treatment. Subsequently, a dental implant was placed and rehabilitated with a provisional crown (Fig. 3). From that point on, follow-up visits and CT scans were conducted annually until the fourth postoperative year.

In the fifth year, the patient did not attend his scheduled follow-up but reported via phone that he had no complaints. During the sixth postoperative year, he was contacted again but failed to return, citing a busy schedule. Four months after this outreach, the patient reported swelling and scheduled a visit. On extraoral examination, a slight, painless increase in volume was observed. Intraoral examination revealed normal mucosal coloration, with no swelling or tenderness upon palpation.

Cone-beam computed tomography revealed an expansive lesion involving the entire region of the iliac graft used in the initial reconstruction. Given the suspicion of tumor recurrence, an incisional biopsy was performed, and histopathological analysis confirmed the recurrence of the primary extracranial meningioma.

In April 2018, a new segmental resection of the mandible was indicated, with 1 cm safety margins. The reconstruction plate was temporarily maintained while awaiting health insurance authorization for either a customized titanium prosthesis or a vascularized bone graft. The surgical approach was carried out via a submandibular incision, allowing exposure of the lesion, followed by resection using a reciprocating saw. The lesion was removed en bloc, including the periosteum surrounding it. The lingual nerve was preserved, as it showed no evidence of tumor involvement, with the lesion confined to the periosteum (Fig. 4).

The patient was advised to maintain a liquid/soft diet until the microvascularized graft reconstruction could be performed. Throughout 2018, he expressed reluctance to undergo reconstruction with a vascularized fibula graft, and the health insurance provider did not authorize reconstructive surgery with a customized plate. In April 2019, the patient returned reporting localized pain and a clicking sensation in the mandibular region. A panoramic radiograph revealed a fracture of the reconstruction plate, which was confirmed by computed tomography, making further delays in the reconstructive procedure unfeasible (Fig. 5).

In May 2019, mandibular reconstruction was performed using a vascularized fibula graft. The surgical approach was again via a submandibular incision, allowing removal of the fractured plate and the condylar segment, which had become unsuitable for plate fixation following tumor resection with safety margins. An additional tumor resection was carried out in the proximal segment using a reciprocating saw, along with debridement of the mandibular canal proximally to the mental foramen and peripheral ostectomy. The lingual nerve was preserved once again, as it showed no evidence of tumor involvement.

The vascularized bone graft was harvested from the left fibula, including the fibular artery and vein, and was dissected and revascularized by an orthopedic surgeon specialized in microvascular techniques. The graft was secured using a right-angle mandibular plate and 2.4 mm locking screws from NeoOrtho (Fig. 6). Surgical closure was performed in anatomical layers.

2.2 POSTOPERATIVE PROGRESS AND FOLLOW-UP

The patient experienced a satisfactory immediate postoperative recovery, with no signs of infection, bleeding, suture dehiscence, or pain complaints. Dental occlusion remained stable, and the patient was discharged on the first postoperative day. On postoperative day 7, he returned for suture removal, reporting mild pain in the fibular donor site but no facial complaints. Moderate edema was still present.

Currently, seven years after the recurrence removal, there is no evidence of tumor recurrence. Imaging studies show proper graft positioning, preservation of bone volume, and maintenance of mandibular contour (Fig. 7). The patient continues to undergo annual clinical and radiographic follow-up for recurrence monitoring and has expressed satisfaction with the treatment received. Cranial CT scans have consistently appeared normal, with no signs of tumor, confirming the primary origin of the lesion in the mandible.

3 DISCUSSION

The theorization surrounding the recurrence of the tumor traces back to the origins of primary extracranial meningiomas. Several mechanisms have been proposed for the development of these lesions: (1) the presence of ectopic meningocytes within the sheaths of cranial nerves as they exit the skull foramina; (2) transformation of undifferentiated mesenchymal cells; (3) displacement of arachnoid cells during embryonic development; and (4) metastasis from an undetected intracranial meningioma. In the present case, given the absence of intracranial involvement and the histological pattern of recurrence, it is plausible to consider the origin of the lesion as either ectopic arachnoid cells or transformed fibroblasts within the mandibular region (Jones, 2001; Mussak, 2007; Landini, 1992; Serry, 2004).

One hypothesis suggests that the inflammatory process triggered by the initial surgery may have stimulated the proliferation of ectopic cells or activated mesenchymal cells (Landini, 1992). Another relevant possibility is that neoplastic remnants may have persisted in the proximal portion of the inferior alveolar nerve (IAN) within the mandibular canal, extending

toward the mental nerve. In the first surgery, the tumor was resected with a 1 cm safety margin based on imaging studies. However, the mandibular body was preserved mesially from tooth 45 onward. This preserved segment may have harbored tumor cells not identified histologically at the time, particularly in the proximal stump of the IAN. Such residual disease could explain the recurrence observed six years postoperatively, as complete lesion removal is one of the most critical factors in preventing recurrence (Simpson, 1987; Budohoski, 2018; Holleczek, 2019).

During the second surgery, which included fibular reconstruction, the mandibular canal was thoroughly debrided proximally to the mental foramen, accompanied by peripheral ostectomy using a carbide "egg burr," thereby reducing the likelihood of residual tumor cells within the proximal canal. Additionally, the possibility of tumor cells persisting in the periosteum cannot be excluded, especially since the recurrence affected the entire region that received the iliac graft—not just the area in contact with the mandibular canal. In the second procedure, the periosteum was dissected and removed along with the lesion, further minimizing the risk of a second recurrence.

Primary extracranial meningiomas may exhibit infiltrative and aggressive behavior despite lacking histopathological features of malignancy, making their clinical course unpredictable (Umana, 2021). Studies report recurrence rates below 20% following complete resection at 5- and 10-year intervals (Rommel, 2017). Nonetheless, rigorous follow-up is strongly recommended—particularly by the European Association of Neuro-Oncology—when bone involvement, mitotic figures, osseous invasion, or necrotic foci are present (Pettersson-Segerlind, 2011; Simpson, 1987; Goldbrunner, 2016). Late recurrence (>10 years) remains a possibility, especially in cases managed with less aggressive approaches such as partial or subtotal resection (Pettersson-Segerlind, 2011). In the present case, recurrence was identified in the sixth year; however, the patient missed the fifth-year follow-up, during which the lesion might have been detected at an earlier stage had it originated from residual IAN involvement. This underscores the importance of continuous monitoring, even in the context of initial clinical stability.

The decision to proceed with microvascularized fibular bone graft reconstruction in response to tumor recurrence was guided by multiple factors. In cases of extensive resections—particularly following failure or limitations of non-vascularized bone grafts, such as the tricortical iliac graft used in the initial surgery—the microvascularized fibular graft stands out for its superior bone repair potential, lower rates of resorption and fracture (Chen, 2019). Additionally, the fibular graft provides adequate bone thickness for future rehabilitation with osseointegrated implants (Manju, 2023). Literature supports this approach especially in

scenarios involving prior reconstructive failure, as demonstrated in the present case (Mertens, 2014; Archual, 2022).

This case report contributes meaningfully to the still limited body of literature on primary extracranial meningiomas, particularly those located in the mandible and treated with microvascular reconstruction following tumor recurrence. While isolated reports do not allow for broad generalizations, the extended follow-up period and the complexity of the surgical approach offer valuable insights into the clinical, therapeutic, and prognostic understanding of this rare entity. The present study underscores the importance of long-term surveillance in such patients and highlights the strategic role of microvascularized fibular grafts as an effective alternative in cases requiring rigorous oncologic control alongside functional and aesthetic rehabilitation. Nonetheless, further studies with larger case series are needed to consolidate treatment protocols and clarify the still poorly understood pathophysiological aspects of primary extracranial meningiomas.

4 FINAL CONSIDERATIONS

This case emphasizes the importance of long-term clinical and radiographic follow-up in patients with primary extracranial meningioma, given its potential for late recurrence—even after initially successful surgical treatment. It also highlights the need for a multidisciplinary approach in complex mandibular reconstructions, particularly in recurrent cases requiring microvascularized bone grafts.

Delays in definitive treatment due to systemic barriers, such as insurance authorization, may lead to complications and necessitate more extensive surgical interventions. Ultimately, this case reinforces that, with proper planning and surgical execution, satisfactory functional and aesthetic outcomes can be achieved—even in challenging reoperative scenarios.

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APPENDICES

Figure. 1 - Panoramic radiograph of the lesion in 2011 affecting the right mandibular branch, with a multilocular appearance at the angle of the mandible

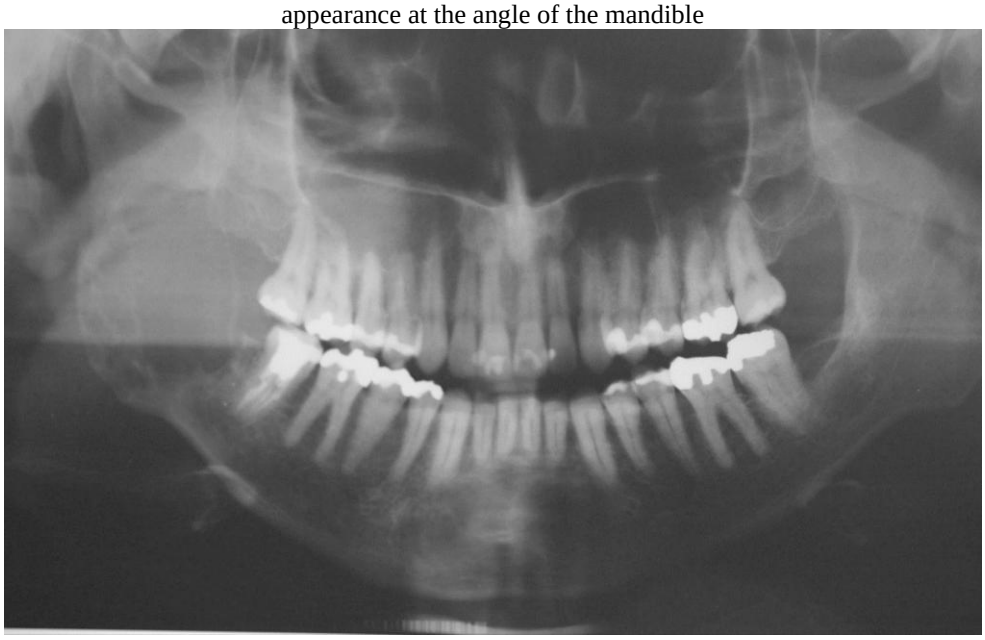

Figure. 2 - Reconstruction with iliac crest graft. Note that the facial artery and vein were preserved.

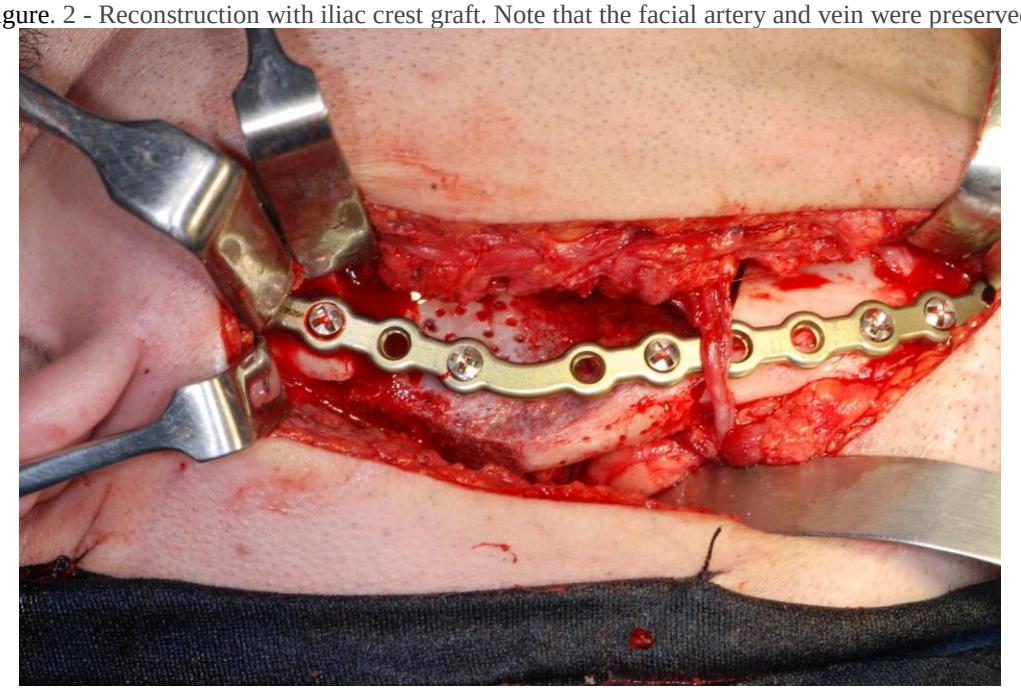

Figure. 3 - Appearance after 18 months of reconstruction with iliac graft and 4 months after implant placement, when the patient received the temporary prosthesis and began orthodontic treatment.

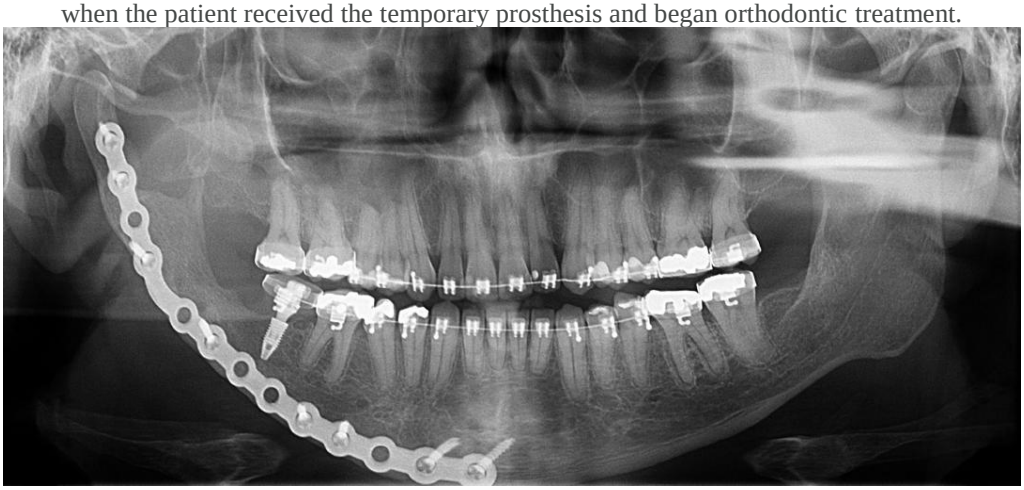

Figure. 4 - CT scan from 2018 after removal of the recurrence tumor. The reconstruction plate was maintained, and the patient was advised to follow a liquid/soft diet until the insurance approved a customized prosthesis or reconstruction with a vascularized fibula.

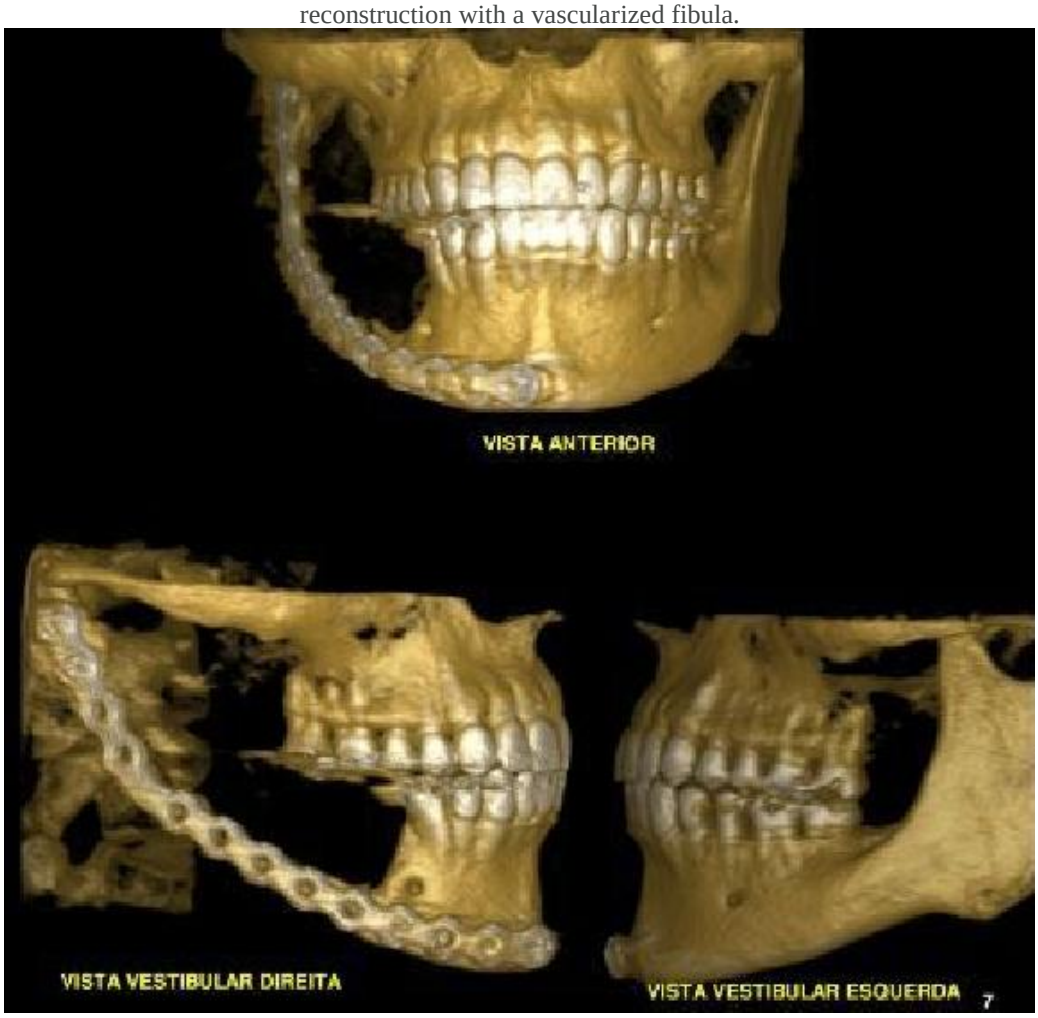

Figure. 5 - Images obtained in 2019 showing the fracture of the reconstruction plate.

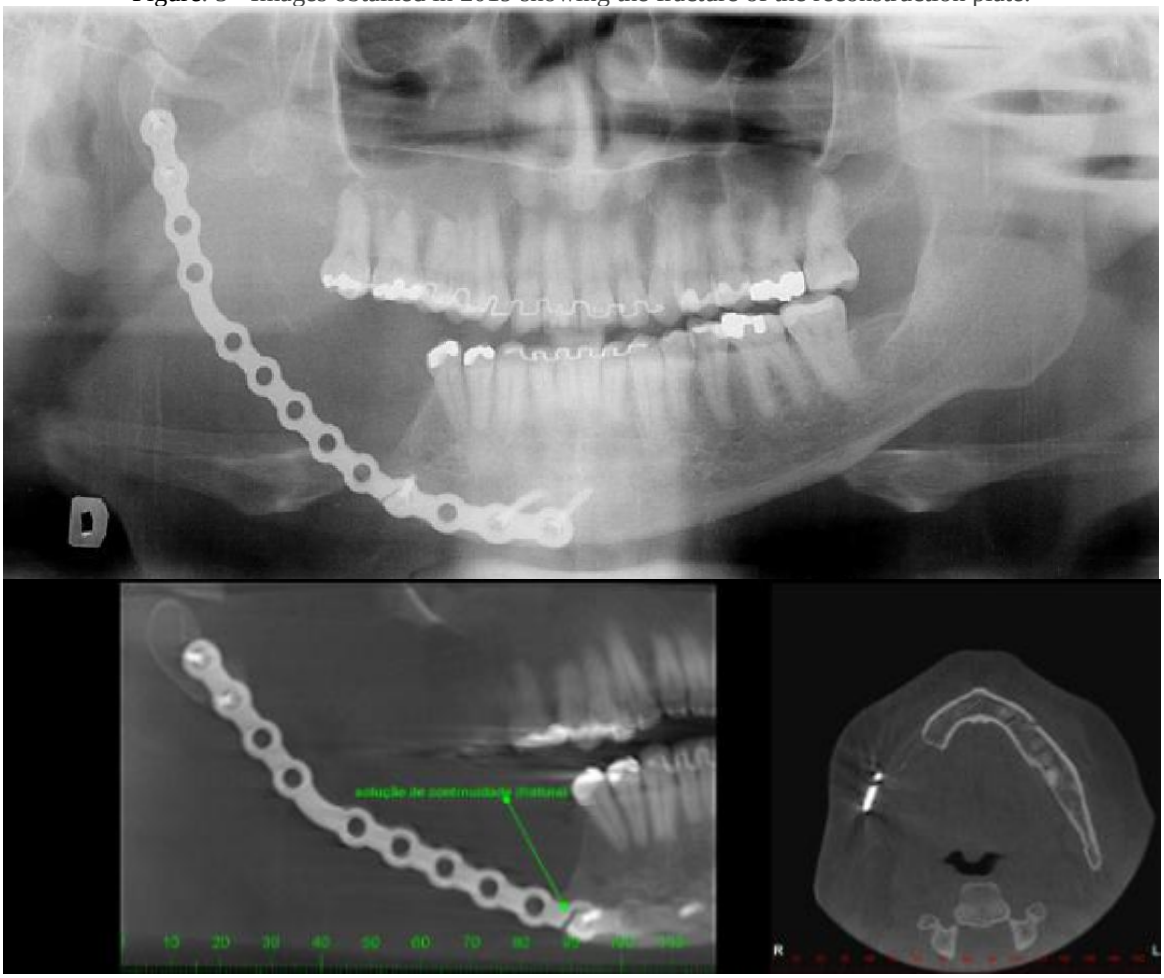

Figure. 6 - Vascularized fibula graft was obtained and adapted to the new reconstruction plate.

Figure. 7 - Images from 7 years of follow-up after removal of the recurrence and reconstruction with vascularized fibula.