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| REVIEW

Etiopathogenesis of pathological mandibular fracture: literature review and case reports

Etiopatogenia da fratura patológica mandibular: revisão de literatura e relatos de caso

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ABSTRACT

Pathological mandibular fractures are fractures induced by pathologies affecting the mandible's base or those resulting from forces that would typically be tolerable if the bone were not compromised by an underlying condition. These fractures often present complex clinical scenarios owing to the compromised bone integrity caused by the pathology, which impedes resolution. Systemic changes may also diminish the capacity for bone neoformation, significantly limiting therapeutic options. Therefore, this study aimed to provide a comprehensive review of the published scientific literature and present clinical cases related to the treatment and prevention of pathological mandibular fractures. A literature review was conducted, focusing on case reports indexed in the MEDLINE and SCIELO databases using specific keywords as descriptors. The search yielded ten articles, which described the etiopathogenesis, classified as infectious, idiopathic, benign pathology, malignant pathology, and iatrogenic. The literature suggests that prioritizing surgery to address the underlying local pathology is crucial. Managing the remaining bone defect optimally may necessitate multiple surgical interventions. Furthermore, preventive measures should be implemented in potential iatrogenic cases. Notably, fractures of malignant, idiopathic, hereditary, and metabolic etiologies may indicate the initial manifestation of diseases.

Indexing terms: Dentistry. Fractures, spontaneous. Mandibular fractures.

RESUMO

As fraturas patológicas mandibulares são aquelas provocadas por patologias que envolvem a base da mandíbula ou que ocorrem por forças de carga que seriam toleradas de forma normal caso o osso não fosse enfraquecido subjacentemente. Os pacientes com fraturas patológicas de mandíbula tendem a apresentar situações clínicas complexas, com limitação ou comprometimento ósseo causado

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pela patologia que dificulta a sua resolução bem como diminuição da capacidade de neoformação óssea pelas alterações de ordem sistêmica que podem restringir significativamente as alternativas terapêuticas. Desta forma, o objetivo do presente trabalho é revisar a literatura científica publicada e apresentar casos clínicos sobre tratamento e prevenção de fratura patológica mandibular. Uma revisão de literatura foi realizada a partir de relatos de caso indexados nas bases de dados MEDLINE e SCIELO com palavras-chave utilizadas de acordo com seus descritores específicos. A pesquisa resultou em dez artigos descrevendo como etiopatogenia causas classificadas como de origem infecciosa, idiopática, patologia benigna, patologia maligna e iatrogenia. A literatura preconiza que a cirurgia para o tratamento da patologia local seja a prioridade e que o defeito ósseo remanescente seja administrado da melhor maneira que o caso permitir, podendo ser corrigido em mais de um tempo cirúrgico. Os possíveis casos iatrogênicos devem ter condutas preventivas. Fraturas de etiologia maligna, idiopática, hereditária e metabólica apresentam a possibilidade de sinalização da primeira manifestação das doenças.

Termos de indexação: *Odontologia. Fratura espontânea. Fraturas mandibulares.*

INTRODUCTION

Pathological fractures of the mandible occur due to conditions that directly impact the mandibular base or from forces that would typically be tolerable if the bone were not compromised by an underlying condition [1]. The decrease in bone strength can be attributed to a variety of factors, including physiological atrophy, widespread benign or malignant diseases, infectious processes, systemic disorders, and medications or treatments that influence bone tissue metabolism. Moreover, surgical procedures like impacted tooth extraction and implant installation can also contribute to bone weakening and subsequent fractures [1].

Pathological mandibular fractures in patients pose intricate clinical scenarios, frequently characterized by compromised bone integrity due to underlying pathology. This compromise impedes effective synthesis and stabilization. Furthermore, a diminished ability for new bone formation and systemic alterations can considerably restrict therapeutic alternatives. Therefore, this study aimed to conduct a comprehensive review of the existing scientific literature and to showcase three clinical cases that highlight the treatment and prevention strategies for pathological mandibular fractures.

Electronic Search

A literature review was conducted, focusing on articles that reported cases of pathological fractures of the mandible. These articles were published between 2013 and 2023 in digital libraries. The databases utilized for this search included the United States National Library of Medicine (MEDLINE; www.pubmed.gov) and the Scientific Electronic Library Online (SCIELO; scielo.org). The search terms were determined using specific descriptors: for MEDLINE (MESH) and SCIELO (DECs). The combinations used were: "pathological fracture [MESH/DECs]" OR "pathologic fracture [MESH]" OR "spontaneous fracture [MESH]" OR "fractures, spontaneous [DECS]" AND "mandible [MESH/DECs]" OR "jaw [MESH]."

Inclusion criteria

Case reports were selected based on the etiopathogenesis of fractures, which included a variety of management approaches documented in the literature. When duplicate pathologies were present, the inclusion criteria prioritized the publication year, with a preference for the most recent article. The diverse causative factors were systematically categorized according to the criteria set by Coletti et al. [1].

CASE REPORTS

The cases reported were categorized based on the same inclusion criteria utilized in the reference articles.

The patients associated with the cases are unidentifiable through the photographs and have provided their signatures on the Informed Consent Form.

RESULTS

The chosen case reports were classified into the following categories: infectious, idiopathic, benign pathology, malignant pathology, and iatrogenic causes. A range of therapeutic strategies were recorded, and comprehensive case descriptions are provided in table 1.

Table 1. Selection of clinical cases according to etiopathogenesis, classification, medical history, therapeutic approach, and relevant study observations.

	Author	Etiopathogenesis	Classification	Medical History	Therapeutic Conduct	Comments
1	Kim et al. [2]	Osteomyelitis	Infectious	Osteoporosis	Affected segment resection and immediate reconstruction with a fibula osteocutaneous flap.	Medication-induced osteonecrosis was part of the differential diagnosis owing to the history of osteoporosis; however, the use of medication was not confirmed.
2	Yoshizawa et al. [3]	Medication-induced osteonecrosis	Iatrogenic	Kidney disease, diabetes, osteoporosis, and heart disease.	Sequestrectomy	The approach was conservative because of the patient's clinical condition. Surprisingly, even without osteosynthesis, the fracture healed, and bone regeneration was observed at the site during the 3-year follow-up.
3	Albarracin-Arjona et al. [4]	Intraosseous hemangioma	Benign Pathology	-	Enucleation of the lesion and intermaxillary fixation.	-
4	O'Brien et al. [5]	Metastatic lesion	Malignant Pathology	Hypertension and gastroesophageal reflux disease.	-	An incisional biopsy performed in the fracture area confirmed the diagnosis of esophageal adenocarcinoma. The patient was staged as advanced and considered inoperable.
5	Tamura et al. [6]	Unifocal Langerhans Histiocytosis	Idiopathic	-	Resection, immediate reconstruction with fibula graft.	-
6	Xiao et al. [7]	Periapical cyst	Benign Pathology	-	Cyst enucleation and osteosynthesis.	-
7	Marunick et al. [8]	Osteoradionecrosis	Iatrogenic	Hypertension, stroke, and head and neck squamous cell carcinoma.	Treatment with hyperbaric oxygenation, resection and immediate reconstruction with fibula graft.	A surgical approach was performed to treat residual trismus from local radiotherapy before the occurrence of osteoradionecrosis.
8	McGoldrick et al. [9]	Impacted teeth removal	Iatrogenic	-	Osteosynthesis	A significant risk of mandibular fracture was evident before the surgery. The fracture occurred 10 days after the extraction, despite the patient being on a liquid and soft diet.
9	Naval-Gías et al. [10]	Periimplantitis	Infectious	Head and neck squamous cell carcinoma	Osteosynthesis	Reconstruction with grafts due to local infection was not indicated in three cases.
10	Ahlers et al. [11]	Traumatic bone cyst	Benign Pathology	-	Curettage, filling of the cystic space with a hemostatic sponge, and osteosynthesis.	-

CASE REPORTS

We discussed three clinical cases pertaining to the treatment and prevention of pathological mandibular fractures. Table 2 illustrates the etiopathogenesis, classification, medical history, therapeutic approach, and pertinent observations of these cases.

Table 2. Etiopathogenesis, classification, medical history, therapeutic approach, and pertinent observations of these cases.

	Etiopathogenesis	Classification	Medical History	Therapeutic Conduct	Comments
Case 1	Osteomyelitis	Infectious	Hypertension and diabetes	Resection of the necrotic area and reconstruction with iliac crest graft.	The reconstruction with an iliac crest graft was performed in a second surgical procedure owing to a local infection.
Case 2	Ameloblastoma	Benign pathology	-	Resection of the lesion and temporary stabilization with a reconstruction plate and surgical cement.	The patient had an associated infectious process, and the reconstruction with an autogenous graft was scheduled for a second surgical procedure.
Case 3	Impacted third molar	Iatrogenic	-	Exodontia and osteosynthesis.	No fracture was noted during the intraoperative period.

CASE 1

A 51-year-old female patient, who is a smoker, reported no clinical issues or medication use. She had a history extraction of tooth 47 owing to mobility, which led to postoperative complications identified as alveolitis. The previous healthcare service treated the alveolitis with curettage and antibiotic therapy. No trauma was reported during or after the procedure. Upon extra-oral clinical examination, an increase in volume was noted on the right hemiface, particularly in the submandibular region. This was accompanied by redness, local warmth, and a fistula. During palpation of the ridge, crepitation and mobility were detected in the mandibular body region.

Panoramic radiography and computed tomography indicated a complex fracture in the right mandibular body (figure 1A-B). During preoperative preparation, the patient was discovered to have uncontrolled hypertension and type II diabetes mellitus. Once the patient's clinical condition stabilized, she underwent a surgical procedure to resect the necrotic area and reduce and fix the fracture. During the operation, areas of bone sequestration were identified and excised. These were sent for histopathological examination, which confirmed a diagnosis of osteomyelitis.

During the 7-month postoperative period, a follow-up imaging examination detected a gap in the mandibular body (not shown), a consequence of residual bone resorption caused by a controlled infectious process. This gap was rectified using a particulate iliac crest graft, which was molded and fixed on-site with fibrin glue. A subsequent check-up, 12 months after this secondary surgical procedure, confirmed graft integration as shown in the control tomography (figure 1D), with no clinical indications of disease (figure 1C).

CASE 2

A 34-year-old female patient reported no clinical issues or medication use. She had previously undergone local curettage at a different healthcare facility, resulting in a histopathological diagnosis of ameloblastoma. The patient exhibited local edema, redness, and warmth, indicative of a secondary infection associated with the tumor. A resection of the mandibular segment was carried out owing to the presence of the tumor. Considering the local infection, the decision was made to conduct rigid osteosynthesis as a temporary reconstruction. This procedure utilized a 2.4 macro plate with angular reinforcement and surgical cement, based on a three-dimensional printed model of the patient's mandible. The goal of this approach was to preserve the soft tissue bed for future definitive reconstruction using autogenous grafts (figure 2A-F).

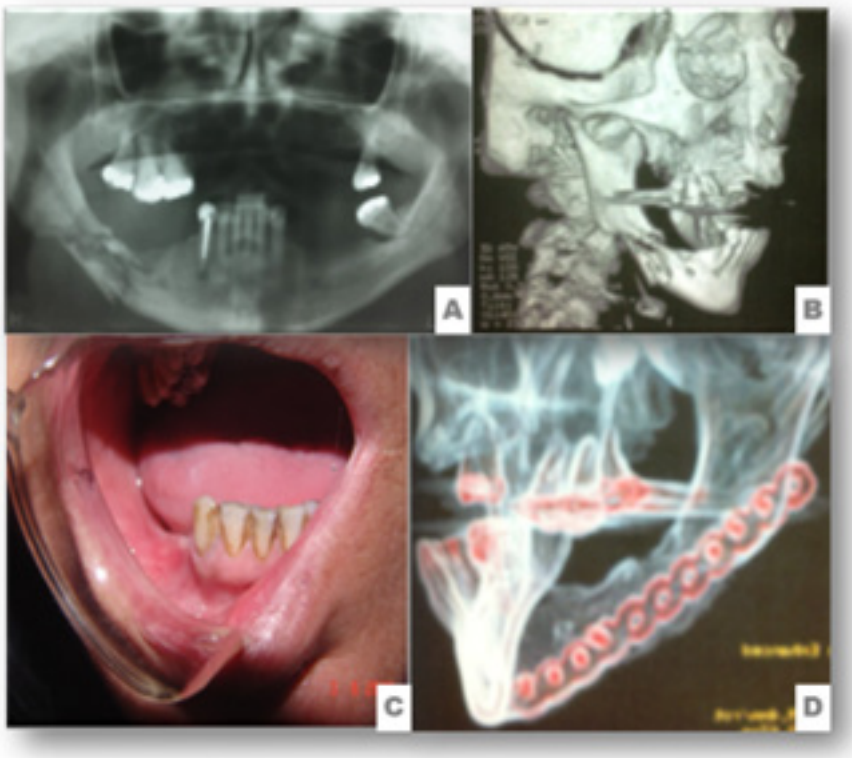

Figure 1. Panoramic radiography (A) and 3D computerized tomography (B) showing the fracture in the region of the right mandibular body. At 12 months post-graft surgery, we observed healing of the mandibular ridge in intraoral view (C). In the oblique lateral view of the computerized tomography, the adaptation of the 2.4 macroplate and integration of the autogenous graft are evident.

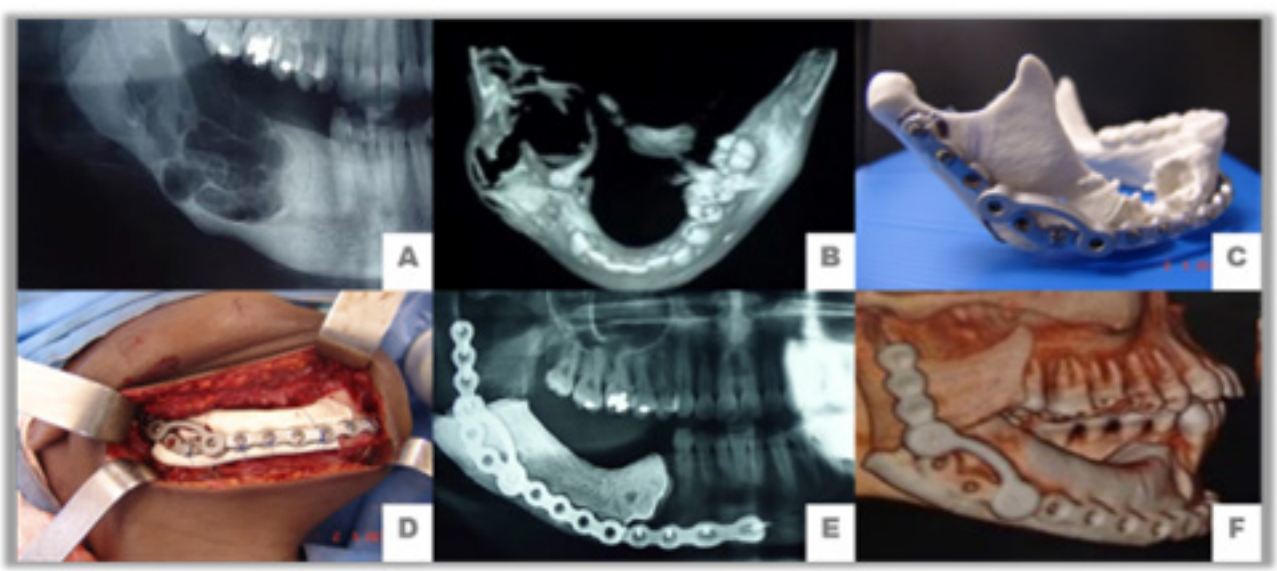

Figure 2. Panoramic radiography (A) and 3D computerized tomography with reconstruction (B) showing the loss of continuity of the mandibular bone structure. In the preoperative phase, prior adaptation of the reconstruction plate with angular reinforcement was performed using prototyping (C). During the intraoperative period, the 2.4 macroplate with angular reinforcement and surgical cement was observed (D). Panoramic X-ray (E) and 3D computerized tomography with reconstruction of the immediate postoperative period (F).

CASE 3

A 51-year-old female patient reported no clinical problems or medication use. She had previously experienced a local infection, which was temporarily resolved through antibiotic therapy. Given the anticipated fragility of the remaining bone and the potential for a mandibular fracture, osteosynthesis was planned following the extraction of the impacted tooth. Although no intraoperative fracture occurred, the osteosynthesis plan was retained as a precautionary measure to prevent a late pathological fracture post-extraction. The histopathological examination of the dental follicle revealed a paradental cyst (figure 3A-C).

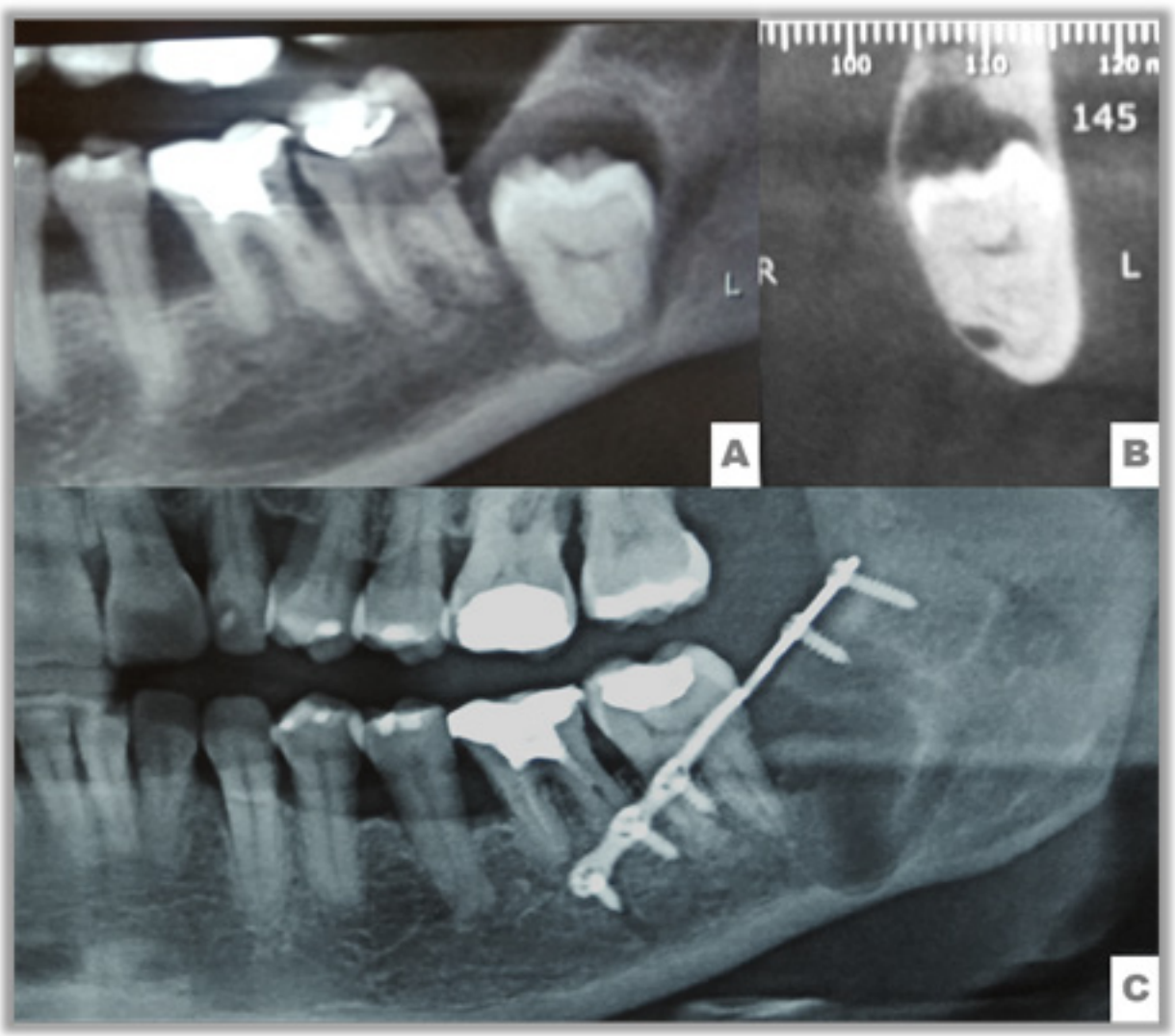

Figure 3. Panoramic radiography (A) and cone beam computed tomography (B) showing the retention of tooth 38 and its positioning in the region close to the mandibular angle. There is a close relationship with the inferior alveolar nerve and limited bone volume in the vestibular, lingual, and basal regions. On the panoramic radiography taken 30 days postoperatively after the tooth extraction (C), the osteosynthesis performed during the intraoperative period is demonstrated, as well as the integrity of the mandibular base.

DISCUSSION

This study aimed to provide a comprehensive enumeration of the various causes of pathological mandibular fractures, covering a broad spectrum of factors. Infectious etiologies identified included periimplantitis [10], while Langerhans-cell histiocytosis [6]. Benign pathologies such as intraosseous hemangioma [4], radicular cyst [7], and traumatic

bone cyst [11] were also observed. Malignant causes were represented by metastasis of esophageal adenocarcinoma [5]. Iatrogenic factors, including osteoradionecrosis [8], medication-induced osteonecrosis [3] and complications from the removal of impacted teeth [9]. Although not documented in this study, it is important to note that hereditary conditions (such as osteogenesis imperfecta), metabolic conditions (osteoporosis), and degenerative conditions (mandibular atrophy) can lead to spontaneous mandibular fractures.

Pathological fracture treatment is complex owing to the variety of etiologies and their influence on bone healing [1]. Consequently, addressing the underlying pathology is paramount, with fracture management contingent on the remaining bone defect [4]. Some authors propose immediate osteosynthesis during pathology treatment, thereby eliminating the need for subsequent reconstruction [10-12]. When necessary, immediate reconstructions using autogenous grafts have been conducted during the initial surgical procedure [6, 8]. In instances of local infection, which heightens the risk of graft loss, delayed reconstructions may be preferred [10]. This strategy was also employed in the clinical cases presented. Moreover, more conservative approaches have been documented, focusing on treating the underlying pathology and achieving mandibular immobilization through maxillomandibular fixation for the fracture [3,4,13].

Case 1 presented a fracture resulting from a tooth extraction complication. The selected treatment was sequestrectomy, reduction, and fixation, as suggested in the literature [6-8,10]. The histopathological diagnosis confirmed osteomyelitis, a condition instigated by Staphylococcus bacteria that rapidly proliferate through the medullary bone, thereby compromising the local blood supply [13]. Preoperatively, the patient was identified with uncontrolled type II diabetes mellitus as a comorbidity, which, when coupled with her smoking habit, heightened her susceptibility to infections [1, 10]. Given the extent of the fracture, osteosynthesis emerged as the sole viable option. However, for cases involving incomplete fractures, only sequestrectomy may be necessary, and the patient can be sustained on a liquid and soft diet for a prolonged period [13]. In instances where the infection area is extensive, segment resection is advised, which can be reconstructed with an immediate graft in the absence of acute infection, as documented [2].

In the treatment of benign pathologies, the focus is on addressing the pathology itself, followed by the stabilization and reconstruction of the mandible based on the remaining bone. Osteosynthesis is performed when conditions permit [12]; however, in certain instances, the affected area is resected and reconstructed with grafts during either the first or second surgical procedure [11]. The second case presented here illustrates the resection of an ameloblastoma and immediate reconstruction using a macroplate and surgical cement. Ameloblastoma, a locally aggressive benign odontogenic tumor, grows by infiltration, distinguishing it from the benign pathologies discussed in this literature review. Despite their benign nature, the inflammatory characteristic of radicular cysts [7] and the vasoproliferative trait of hemangiomas result in different disease progression patterns, thereby justifying more or less radical surgical approaches.

Iatrogenic pathological fractures can result from osteoradionecrosis, medication-induced osteonecrosis, the removal of impacted teeth, and implant installation [1]. While these procedures can be planned and delayed to prevent fractures when elective, the onset of osteoradionecrosis and medication-induced necrosis may sometimes be inevitable, prompting a reconsideration of this classification. The literature has reported instances of mandibular fractures caused by osteoradionecrosis during surgical intervention in a previously irradiated area. This procedure was necessitated by severe trismus induced by the radiotherapy itself, with the goal of preventing nutritional deficiencies that could exacerbate the patient's prognosis [8]. Similarly, a case of spontaneous medication-induced osteonecrosis was reported, diagnosed based on the patient's clinical manifestation, with no prior surgical manipulation of the affected area. Furthermore, the oral environment was deemed adequate before the initiation of drug therapy, yet osteonecrosis developed 2 years later [3].

The prediction of areas with diminished bone support following the extraction of impacted teeth, which are unable to endure masticatory stresses during bone consolidation phases, necessitates additional precautions. These include extending the consumption of liquid and soft diets for a minimum of 4 weeks postoperatively [13]. This raises the question of whether other preventive measures could be implemented during the intraoperative period, thereby classifying such occurrences as iatrogenic [9]. The placement of osteosynthesis plates, even in the absence of an intraoperative fracture, presents a viable alternative for maintaining the integrity of the weakly structured mandibular base post-surgery. This contributes to the prevention of post-operative pathological mandibular fractures, as illustrated in Case 3.

Pathological fractures have been reported to potentially correlate with idiopathic diseases such as Langerhans cell histiocytosis. This term refers to a wide range of diseases that result in bone destruction and share similar histological characteristics. However, these diseases present at varying age peaks and locations, and they carry different prognoses. This review discussed a case of unifocal disease; however, it is crucial to continually monitor the patient. This is because multifocal manifestations of the condition are linked with high recurrence and mortality rates [6].

Although exceedingly rare, it has been observed that pathological fractures in the mandible can result from distant metastases of malignant neoplasms. Bone metastases in the facial region are infrequent, yet when they do occur, they represent 25% of primary tumor diagnoses, often developing without symptoms [5]. This underscores the critical role of histopathological examination, not only in instances of spontaneous fractures but also in various other oral and maxillofacial surgical procedures, given the potential systemic implications of facial lesions.

The current study aimed to explore the causes and treatment strategies for pathological fractures of the mandible, a rare condition that accounts for less than 2% of all mandibular fractures. Despite their rarity, these fractures present a significant challenge in the field of oral and maxillofacial surgery. While benign pathologies can be swiftly addressed based on their nature, causes such as idiopathic, malignant, metabolic, hereditary, and degenerative conditions require a more thorough investigation of the patient's systemic health. Iatrogenic causes highlight the necessity for preventive measures tailored to this specific patient profile. Additionally, it is important to note that infectious processes may be either secondary to other conditions or independent, a distinction that is critical when deciding on immediate graft reconstructions due to the increased risk of complications.

CONCLUSION

The prevailing consensus in the literature suggests prioritizing surgical intervention to address the root pathology, with subsequent management of any residual bone defect. Immediate correction should be implemented when suitable. In instances where patients with spontaneous fractures lack optimal systemic conditions or viable bone remnants, a secondary surgical reconstruction is a safer alternative that can mitigate postoperative complications. Iatrogenic cases, despite their potentially contentious classification, should be managed proactively, employing strategies to prevent fracture incidences whenever feasible.

Thorough investigations should be conducted on fractures of malignant, idiopathic, hereditary, and metabolic etiology, as they could potentially be the initial manifestation of these diseases. This underscores the significance of precise diagnosis, meticulous planning, and individualized treatment strategies for pathological mandibular fractures, with the goal of enhancing prognosis and patients' quality of life.

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