

Original Article

Pathological Fractures Of The Jaws Due To Cystic Lesions: A Three Case Series With A Brief Review Of The Literature

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Abstract

This study aims to describe pathologic fractures of the jaws due to cystic bone lesions. Their daily clinical practice findings are not very common, so their treatment can be challenging. In addition, endosseous benign lesions like a radicular, residual, solitary, aneurysmal cyst, and odontogenic keratocyst, might weaken the bone so that trauma, or usual chewing, could break it. We follow from the first clinical examination to the X-ray or CBCT examination and surgical management of our patients, reporting all procedures and results to explain our approach to these cases. We also reviewed the literature briefly to determine if our operative management is in line with the scientific community. Mandibular angle and body are common locations, while symphysis and condyle are less so. Young men are the most affected, and trauma is the most triggering event. Cyst enucleation, followed by fracture reduction and fixation, is the treatment of choice for our clinical team and the scientific community. We observe how impactful it is to approach the cavity of the cystic lesion from the fracture line as the surgical gold standard.

Keywords: *Pathological fractures, cystic lesions, maxilla, mandible.*

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Introduction

The occurrence of pathological fractures of the jaws due to benign cystic lesions is rare. The weakening of the bone that leads to fracture, either during normal function or consequent to trauma, is mostly associated with follicular cysts, residual and radicular cysts, aneurysmal cysts, and odontogenic keratocysts⁽¹⁻¹⁰⁾. This paper aims to describe three case reports we came across between 2018 - 2020 and evaluate the literature about etiology, site frequency, sex and age distribution, and prevalent treatment.

Case presentation

Case number 1

In 2018, 54 years old, in good health, the female was referred to the Oral Surgery Unit of the School of Dental Sciences (University of Trieste, Italy) due to an accidental fall that caused her a maxillo-zygomatic complex fracture. Besides its usual pattern, we detected with the CBCT (MyRay Hyperion X9 with FOV from 5x5 to 11x5 mm, sagittal view from section 1 to 39) a 22,5x28,8 mm intrasinus and unilocular cystic lesion, expanding to the anterior and lateral right maxillary sinus walls. The apices of the second upper right premolar and the first molar were inside the hypodense lesion; both teeth had deep cavities with no response to vital pulp tests. The patient underwent surgery to reduce and fixate the fracture under general anesthesia (Fig. 1a and 1b). An intraoral approach was performed, reaching the lesion entering the fracture gap, and involved teeth were extracted. The anterior maxillary sinus wall, between the zygomaticomaxillary and the nasomaxillary buttresses, was crossed by a horizontal fracture running over the enucleated cystic lesion. The zygomaticomaxillary complex fracture was reduced and fixed in a three-point way being the sinus walls the last ones in the sequence. The alveolar mucosa was watertight and sutured in a two-layer fashion, sliding a buccal fat pad flap to minimize the risk of developing an oroantral fistula.

Histopathologic examination depicted the lesion as a radicular cyst.

Case number 2

In the summer of 2019, 19 years old, in good healthy male was referred to our Unit with left parasymphiseal and left undisplaced incomplete mandibular angle

fractures reported after being involved in a fight. Diagnostic insights with CBCT (MyRay Hyperion X9 with FOV from 5x5 to 11x5 mm, sagittal view from section 61 to 83) scans showed a vertical linear symphyseal fracture line along an unrecognized, silent, asymptomatic, hypodense bone lesion measuring 2 cm in its mesiodistal diameter, 1 cm in height and 1,5 of width. The teeth 3.3, 3.2, 3.1, 4.1, and 4.2, whose apices were radiographically over-imposed to the lesion, were revealed to be positive to the vital pulp test. The patient reported he had been a victim of facial injuries in the past, both for sports activity and brows. He was put into an elastic intermaxillary fixation on orthodontic brackets before undergoing surgery under general anesthesia. A vestibular cortex window was performed, and the lesion was approached. No capsule lined the wall, so it was gently scratched. Blood was found inside the lesion, with little debris collected for histopathologic examination just before fixing the fracture along the tension lines of the symphysis and repositioning the vestibular bone window. (Fig. 2a and 2b). The histological result showed a polychrome fibroblastic connective tissue with giant cells. Given the history of the patient and the exam result, a TBC (Traumatic Bone Cyst) diagnosis has been made.

Case number 3

A 86 years old, in good health female patient with no history of recent trauma was hospitalized in 2020 due to a painful hard swelling along the left mandible body that slowly developed within the last two weeks. Intraorally, a fixed partial prosthesis badly rested over residual roots and hopeless teeth. A purulent fistula was present on the alveolar crest close to the left lower canine, refractory to antibiotic therapy. The orthopantomography showed an atrophic mandible, with a left parasymphiseal vertical fracture superimposed to a round radiolucency around the left lower canine tooth. Echography revealed multiple Ia and Ib lymph nodes swelling with benign inflammatory patterns. The CT scans (MyRay Hyperion X9 with FOV from 5x5 to 11x5 mm, sagittal view from section 46 to 71), with a contrast agent, confirmed the benign origin of the bone lesion. (Fig. 3a and 3b). Under general anesthesia, the fracture has been approached submentally. Lymph nodes of I station, granulation, and inflammatory tissue inside the fracture site have been removed and sent for the histopathological exam. Due to the severe atrophy, the mandible fracture has been fixed with a load-bearing 2.5 MODUS® Trilock titanium plate (Medartis, Switzerland) pre-plated extra orally over a 3D printed model from an STL file of the mirrored right healthy side.

The histopathological exam revealed lymph nodes histiocytosis with plasma cells infiltrating and cystic tissue with necrosis and inflammation (inflammatory cyst).

All case reports presented have a CT as the first radiological exam. However, the nature of the post-surgery exam is different, specifically in observing different parts of maxillary bones. We also considered it appropriate during the Covid epidemic to restrict access to our operative Unit, so we did not perform new and homologous radiographic exams on our patients.

Table 1: Inclusion and exclusion criteria.

| Inclusion criteria | exclusion criteria |
|---------------------------|--|
| English language | Other languages |
| All paper types | Fracture subsequent to |
| No age cut-off on samples | Difficulties in the availability of papers |
| No sex cut-off on samples | |

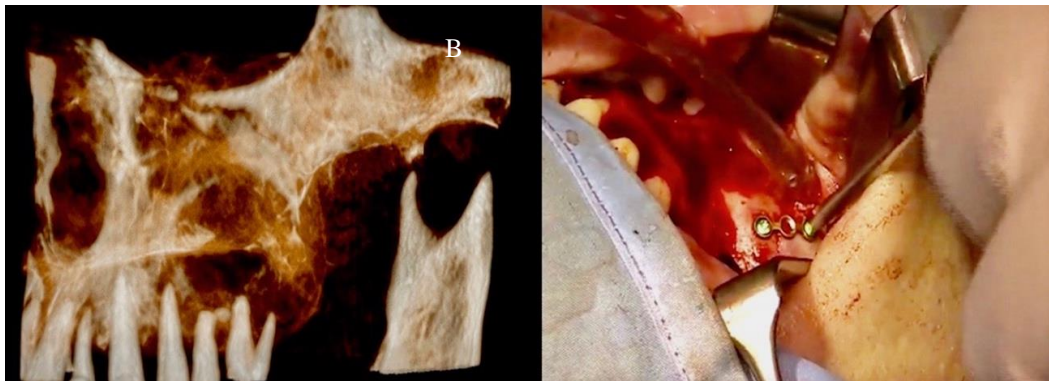


Figure 1: (A) A 3D reconstruction by CBCT of case 1. (B) Bone fixation after removing the cystic lesion in case 1.

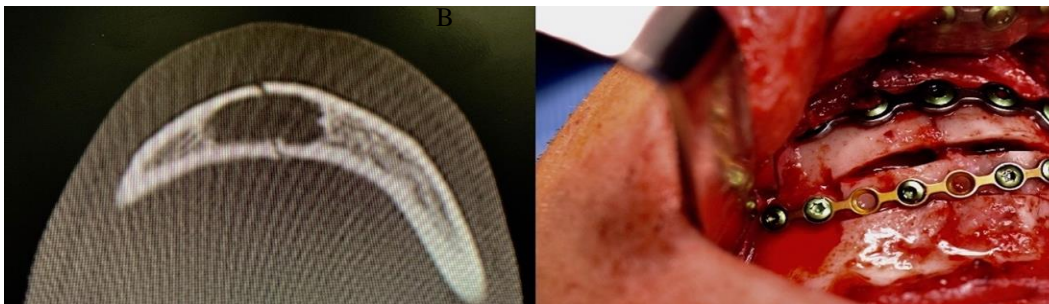


Figure 2: (A) Evidence of line fracture on CBCT of case 2. (B) Bone fixation after removing the cystic lesion in case 2.

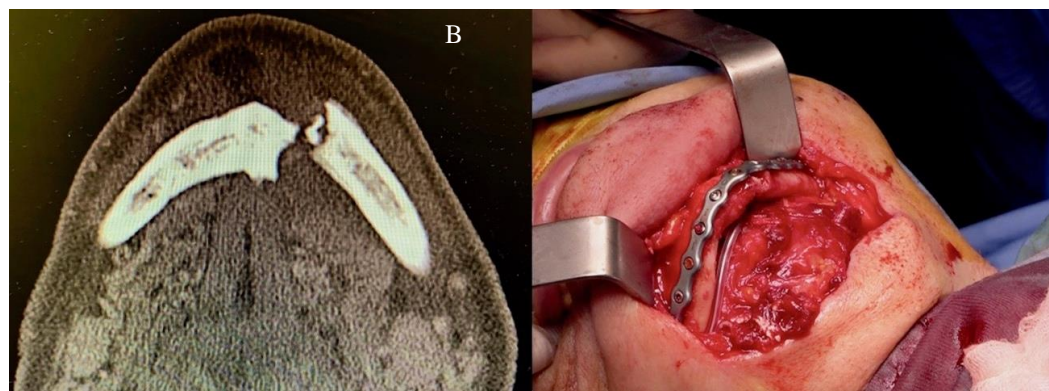


Figure 3: (A) Evidence of line fracture on CBCT of case 3. (B) Bone fixation after curettage and reduction of the fracture in case 3.

Table 2: Table of articles included in the systematic review.

| Authors | Publication | Paper type | Site | Lesion's type | M/F | Treatment |
|-----------------------|-------------|------------|-----------------------|----------------------|-------------|------------------------------|
| Grether et al. | 1951 | CR | MANDIBLE | ODONTOGENIC CYST | | SURGERY |
| Martensson et al. | 1957 | CR | MANDIBLE | ODONTOGENIC CYST | M,50 yo | SURGERY (from the gap) |
| Baird et al. | 1958 | CR | MANDIBLE | SBC | M,19 yo | SURGERY |
| Marsden et al. | 1964 | REVIEW | MANDIBLE | ODONTOGENIC CYST | 2 CASES | SURGERY |
| Zambito et al. | 1964 | CR | MANDIBLE | ODONTOGENIC CYST | | |
| Matise et al. | 1967 | CR | MANDIBLE | TRAUMATIC CYST | | SURGERY |
| Jablonski et al. | 1967 | CR | MANDIBLE | ODONTOGENIC CYST | | SURGERY (from the gap) |
| Ezsias et al. | 1994 | REVIEW | | | 4 CASES | |
| Gerhardes et al. | 1998 | REVIEW | | | | |
| Motamedi et al. | 1998 | REVIEW | MANDIBLE | ODONTOGENIC CYST | | SURGERY |
| Amos et al. | 2007 | CR | MAXILLA | ODONTOGENIC CYST | M, 20 yo | SURGERY |
| Goddard et al. | 2007 | CR | MANDIBLE | ANEURISMATIC CYST | | SURGERY |
| Coletti et al. | 2007 | REVIEW | | | | |
| Choi et al. | 2011 | CR | MANDIBLE (Condyle) | ANEURISMATIC CYST | | SURGERY (double approach) |
| Abir et al. | 2013 | REVIEW | | | | |
| Boffano et al. | 2013 | REVIEW | | | | |
| Ahlers et al. | 2013 | CR | MANDIBLE | TRAUMATIC CYST | M,13 yo | SURGERY |
| Kouhsoltaui et al. | 2015 | CR | MANDIBLE | ODONTOGENIC CYST | M,38 yo | SURGERY |
| Chell et al. | 2017 | CR | MANDIBLE | SBC | M,16 | SURGERY |
| Xiao et al. | 2018 | CR | MANDIBLE | ODONTOGENIC CYST | F,49 yo | SURGERY |

Discussion and conclusions

Our clinical practice on pathological fractures is weekly, but we need to deepen our knowledge on the subject. The brief review of the literature performed started researching for “cyst jaw fracture”, “pathological jaws

fracture cyst”, “pathological fracture mandible,” and “pathological fracture maxilla” as keywords on PubMed from inception to June 2021. According to our inclusion and exclusion criteria (Table 1), just 20 of the 150 papers found have been selected (Table 2). The case report was the most common feature, maybe due to the lack of

frequency in everyday practice. Reviews of literature come just behind.

Even though the literature has always given ample space to pathological fractures, the oldest article is dated 1951⁽¹⁾; one of the most significant reviews was drawn up by Boffano et al. in 2013.

In this paper, authors considered multiple determinant causes for pathological fractures: tooth removal implant placement or periimplantitis, malignant pathologies or benign tumors, Osteomyelitis or Osteoradionecrosis (ORN), and Biphosphonate Related Osteonecrosis of the Jaws (BRONJ)⁽²⁻⁵⁾.

Pathological fractures of the jaws are rare. Gerhards et al. state that pathological fractures of the mandible are less than 2% of the incidence of all fractures. In Boffano et al. paper, the incidence is the state of being higher in men, and the ratio of male to female changes according to the different factors determining the fracture: is 2.2:1 for pathological fractures caused by tooth removal, 1:3 when it is secondary to implant placement, 1:1 for pathological fractures depending on tumors. In the case of a benign cyst, the incidence is higher in males.

The mandible is the most common site where pathological fractures develop^(1-6,9-13). In particular, Gerhards et al. found the molar site, the mandible's angle, and the condyle to be the most affected sites by pathological fractures of the jaws⁽⁵⁾. On the other hand, Boffano's et al. review distinguished mandibular symphysis as the most common site of fracture in edentulous patients and the other parts of the mandible for cystic lesions or benign and malignant tumors instead⁽²⁾.

What caught our attention was that a part of these pathological fractures is due to cystic lesions not recognized up to the fracture. Furthermore, literature pays particular attention to pathological fractures caused by Traumatic Bone Cysts, named like this according to the WHO⁽¹⁴⁾, otherwise known as Hemorrhagic Bone Cysts or Solitary Bone Cyst or Idiopathic Bone Cyst. Although it rarely occurs in the jaw (1% of its localization, being the long bones the most common)^(4,7,12-14), it is well represented in literature⁽⁷⁻¹⁹⁾.

Odontogenic and TBC as the cystic patterns most frequently associated with pathological fractures of the maxillary bones^(2,5-15,22,23). Age is not a significant factor for developing this type of fracture, as odontogenic cysts are more frequent in patients during their III – V decades of life. Still, on the other hand, a lively and sporty lifestyle, not only at a young age, can promote the development of TBC^(9-13,15-16).

Patients affected by pathological fractures due to benign cysts have a mean age of 43.75 years, ranging between 16 and 76 years^(2,4,5-11). It may occur in elderly patients due to their predisposition to malignant disease and atrophic edentulous jaws⁽⁵⁾.

The treatment of choice is the contextual enucleation of the cyst and reduction of the fracture following the rationale to improve postoperative morbidity and prognosis^(11-19,24-28).

The cases we presented confirm this trend. Out of three exposed cases, in two mandibles was the affected site, and the most frequently associated cystic lesion pattern is odontogenic. However, having also found a TBC, it is possible to state that this particular cystic pattern, although rare, has its relevance.

In 1982 Cope described a pathological fracture in an atrophic mandible treated without fixation and concluded that this could be good for atrophic bones, a simple fracture with minimum displacement in elderly and infirm patients. But following literature agree that internal fixation is the treatment of choice in all pathological fractures of the jaws. Therefore, approaching patients in their entirety and with strong planning for the surgery with the right specificity, the *modus operandi* mostly approved in literature is followed. We considered the gap fracture the right access route for cyst enucleation and fracture reduction and fixation in all cases. The enucleation of the cystic lesion before and afterward fracture reduction and fixation with osteosynthesis screws and plates is the gold standard we decided to follow. Using these procedures in all our three cases, follow-ups at 1, 7, and 21 days after surgery were positive and notable improvements were recorded regarding symptoms and function, which was our first required goal.

Traditional open reduction and internal fixation associated with cyst enucleation is the treatment of choice⁽¹¹⁾. However, when the bone deficit is so wide, many authors agree with resectioning the diseased area and promoting an immediate reconstruction. Chell et al.⁽³⁾ point out as the fracture gap might be the right access to the cyst enucleation. It is also worth mentioning an alternative treatment method with a combination of a reconstruction plate, recombinant human bone morphogenetic protein 2, and tricalcium phosphate⁽³⁰⁾ or just with an iliac crest bone graft⁽³¹⁾.

It is possible to state that bone deficit caused by cystic lesions can be so great as to lead to pathological fracture of the jaws spontaneously (in severe atrophy) or traumatically. In both cases, the treatment is complicated due to the reduced quantity of healthy tissue in correspondence with the abutment's faces and the possible consequent instability of the fixation. Accurate diagnosis, knowledge of biomechanics and occlusal forces, virtual planning and choice of the correct hardware, and an appropriate surgical technique are key elements in these clinical conditions. In extreme tissue deficits, therapeutic success can be achieved through bone grafts which therefore find a very appropriate indication.

Our clinical practice is studded with cases of pathological fractures of the jaws. Therefore, the literature review was useful in understanding which type of cystic lesion is the most frequently associated with pathological fractures, as previously mentioned, the odontogenic pattern, and the correct way to treat them. Also, in our daily practice, TBC is a reality to be considered when formulating diagnostic suspicion. Therefore, we approach patients entirely, taking into account their specificities (age, sex, medical history, and cause of the fracture) while relying on the same surgical procedures.

To achieve the main aims, it is important to plan the correct treatment and implement effective surgical techniques, when needed, with bone grafts to fill the residual bone deficit.

References.

- Grether A, Vidal P, Meyer J. Paradental cyst of the mandible evolving into a fracture. *Inf Dent*. 1951;33(6):295-300.
- Boffano P, Rocchia F, Gallesio C, Berrone S. Pathological mandibular fractures: a review of the literature of the last two decades. *Dent Traumatol* 2013;29(3):185-96.
- Chell M, Idle M, Green J. Case Report: An unusual finding of a solitary bone cyst in a patient with a fractured mandible. *Dent Update*. 2015;42(10):977-78.
- Choi BJ, Choi SC, Kwon YD. Aneurysmal bone cyst causing a pathologic fracture of the mandibular condyle. *J Oral Maxillofac Surg*. 2011;69(12):2995-3000.
- Gerhards F, Kuffner HD, Wagner W. Pathological fractures of the mandible. *Int J Oral Maxillofac Surg*. 1998;27(3):186-90.
- Ezsias A, Sugar AW. Pathologic fractures of the mandible: a diagnostic and treatment dilemma. *Br J Oral Maxillofac Surg*. 1994;32(5):303-6.
- Kalantar Motamedi MH. Aneurysmal bone cysts of the jaws: clinicopathological features, radiographic evaluation and treatment analysis of 17 cases. *J Craniomaxillofac Surg*. 1998;26(1):56-62.
- Goddard R, Patel N. Aneurysmal bone cyst masquerading as unknown mandibular metastatic deposit causing pathological fracture. *Dent Update*. 2007;34(4):230-2.
- Madiraju G, Yallamraju S, Rajendran V, SrinivasaRao K. Solitary bone cyst of the mandible: a case report and brief review of literature. *BMJ Case Rep*. 2014;bcr2013200945.
- Xiao X, Dai JW, Li Z, Zhang W. Pathological fracture of the mandible caused by radicular cyst: A case report and literature review. *Medicine (Baltimore)*. 2018;97(50):e13529.
- Coletti D, Ord RA. Treatment rationale for pathological fractures of the mandible: a series of 44 fractures. *Int J Oral Maxillofac Surg*. 2008;37(3):215-22.
- Hs CB, Rai BD, Nair MA, Astekar MS. Simple bone cyst of mandible mimicking periapical cyst. *Clin Pract*. 2012;2(3):e59.
- Velasco I, Cifuentes J, Lobos N, San Martín F. The unusual evolution of a simple bone cyst in the mandible: A case report. *J Clin Exp Dent*. 2012;4(2):e132-5.
- Kramer IR, Pindborg JJ, Shear M. The WHO Histological Typing of Odontogenic Tumours. A commentary on the Second Edition. *Cancer*. 1992;70(12):2988-94.
- Saito Y, Hoshina Y, Nagamine T, Nakajima T, Suzuki M, Hayashi T. Simple bone cyst. A clinical and histopathologic study of fifteen cases. *Oral Surg Oral Med Oral Pathol*. 1992;74(4):487-91.
- Harnet JC, Lombardi T, Klewansky P, Rieger J, Tempe MH, Clavert JM. Solitary bone cyst of the jaws: a review of the etiopathogenic hypotheses. *J Oral Maxillofac Surg*. 2008;66(11):2345-8.
- Lucas CD, Blum T. Do all cysts in the jaws originate from the dental system?. *J Am Dent Assoc* 1929;16(4):647-61.
- Howe GL. Haemorrhagic cysts of the mandible. *Br J Oral Surg*. 1965;3(1):55-76.
- Jabłoński M. Treatment of the mandibular fractures with the course through the lumen of the odontogenic cysts. *Czas Stomatol*. 1967;20(4):363-8.
- Marsden J. Fracture of the mandible due to radicular and residual odontogenic cysts. *Br J Oral Surg*. 1964;2(5):71-5.
- Amos M, Dalghous A, Alkhabuli J, Mizen K. Massive maxillary radicular cyst presenting as facial fracture and abscess, a case report. *Libyan J Med*. 2007;2(4):211-3.
- Baird WO, Askew PA. Traumatic mandibular bone cyst involved in line of fracture. *Oral Surg Oral Med Oral Pathol*. 1958;11(12):1351-6.
- Martensson G. Bone cysts of the mandible. *Oral Surg Med Oral Pathol*. 1965;19:639-54.
- Zambito RF, Laskin DM. Follicular cyst of mandible associated with pathologic fracture: report of a case. *J Oral Surg Anesth Hosp Dent Serv*. 1964;22:449-52.
- Matise JL, Beto LM, Fantasia JE, Fielding AF. Pathologic fracture of the mandible associated

- with simultaneous occurrence of an odontogenic keratocyst and traumatic bone cyst. *J Oral Maxillofac Surg.* 1987;45(1):69-71.
26. Ahlers E, Setabutr D, Garritano F, Adil E, McGinn J. Pathologic fracture of the mandible secondary to traumatic bone cyst. *Craniofac Trauma Reconstr.* 2013;6(3):201-204.
 27. Abir B, Guerrouani A, Abouchadi A. Pathological fractures of the mandible: A report of ten cases and a review of the literature. *Open J Stomatol.* 2013;3(8):419-24.
 28. Kouhsoltani M, Mesgarzadeh A, Moradzadeh K. Mandibular fracture associated with a dentigerous cyst: report of a case and literature review. *J Dent Res Dent Clin Dent Prospects.* 2015;9(3):193-8.
 29. Cope, M. Spontaneous fracture of an atrophic edentulous mandible treated without fixation. *Br J Oral Surg.* 1982;20(1):22-30.
 30. Castro-Núñez J, Cunningham LL, Van Sickels JE. Atrophic Mandible Fractures: Are bone grafts necessary? an update. *J Oral Maxillofac Surg.* 2017;75(11):2391-8.
 31. Alagöz MS, Uysal AC, Sensoz O. An alternative method in mandibular fracture treatment: bone graft use instead of a plate. *J Craniofac Surg.* 2008;19(2):411-20.