Conservative Management of Mandibular Odontogenic Myxoma: Report of Two Cases

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Abstract

Odontogenic myxoma is a slow growth, locally aggressive benign tumor of ectomesenchymal origin. Although rare and unencapsulated, commonly shows similar characteristics to other disorders of the jaws. Clinically it can be asymptomatic, and affect dental and nervous structures. The diagnosis is based on imaging and histopathological examinations. Treatment has no surgical guidelines and many varied approaches, based on the lesion size and location.

The recommended treatments, as surgical resections with safety margin and curettage associated with alternative methods, aim to minimize relapse. There is no predictive prognosis for this tumor and each case must be individually evaluated. This paper aims to exemplify important aspects to be considered in guiding the therapeutic approach, reporting two cases in adolescent patients. The first case was treated by resection aided by a custom 3D-printed osteotomy guide, while the other underwent curettage with cryotherapy, and no recurrence evidence in 4 and 5-year follow-up, respectively.

Keywords: Odontogenic myxoma • Cryotherapy • Odontogenic tumors • 3D-printed guide

Introduction

Odontogenic Myxoma (OM) is a rare benign tumor, representing from 3 to 5% of odontogenic tumors and can lead to high relapse rates after treatment. It has an ectomesenchymal origin and is not encapsulated. This tumor usually affects the mandibular bone of young adults, with no gender predilection. It constitutes a heterogeneous group with different histopathological characteristics and several clinical manifestations, which, despite being considered benign, is locally aggressive, slow-growing, and can reach considerable size [1]. In addition to cortical bone expansion, it can cause tooth displacement and occlusion changes, but root resorption is rarely observed [2].

Regarding clinical examination, these tumors produce few symptoms, presenting a firm expansive mass in the maxillomandibular complex, with or without displacement and/or mobility of the associated teeth. More developed lesions might present signs and symptoms such as swelling caused by bone expansion, pain, paresthesia, and damage to dental elements close to it or contained in the lesion [3]. Odontogenic myxomas more frequently affect the mandible body and angle regions as well as the ascending ramus. When the zygomatic bone and the maxillary sinus are affected, a more severe condition is observed due to their destruction power.

Their imaging characteristics include unilocular or multilocular radiolucent lesions, and several terms have been used to describe these characteristics such as “soap bubble” and “honeycomb” aspect [4]. However, none was considered specific of these lesions due to the variety of images produced and similarity between its differential diagnoses [5]. Due to their clinical and imaging similarity with other maxillary lesions, it is necessary to resort to the differential diagnosis to prevent the use of an unsuitable treatment. According to the imaging aspects, the OM differential diagnosis includes the following lesions: Ameloblastoma, aneurysmal bone cyst, intraosseous hemangioma, cherubism, giant cell lesion, keratocyst, and osteosarcoma [6].

Histological diagnosis is the key to selecting the treatment modality and must be done as soon as possible to prevent expansion of the jawbones and need of resection or cause pathologic fractures.

Their histological features include fusiform, round, angular cells embedded in abounding mucoid stroma or randomly arranged in a loose myxoid containing only some collagen fibrils, in the presence or absence of odontogenic epithelium islands [7]. Many theories support the OM odontogenic origin due to the histological similarity between the lesions and the pulp ectomesenchyme, constant association with missing or not erupted teeth and occasional presence of inactive odontogenic epithelium, as well as its unusual occurrence in other parts of the body. Small lesions are usually treated with curettage provided that a thorough follow-up of at least 5 years is kept. However, larger lesions require complete removal with a safety margin, segmental resections or hemi-mandibulectomy followed by bone reconstruction and a multidisciplinary team for rehabilitation [8,9].

In general, the prognosis is favorable; however, relapse might occur. The mean rate of recurrence is 25% and can be depending on the treatment, but usually occurs within two years. The recurrence seems to decrease, irrespective of whether resection or a more conservative approach is used, after 10 years of surgical intervention. However, this tumor does not present malignant potential.

Conservative treatments might include enucleation, curettage, and marginal resection and might be considered in younger patients.

Cryotherapy is a technique applied as an ancillary in conservative treatments. Its principle is based on tissue destruction by freezing, which is called cryolesion. Decreased tissue temperature results in the formation of ice crystals in the extracellular medium, which increases the electrolyte concentration. This creates a hyperosmotic

hyperosmotic environment that results in water osmosis outside the cells generating increased concentration of intracellular electrolytes, pH alteration, and protein denaturation. After defrosting, a new cooling provokes the formation of ice crystals in the intracellular medium. When the ice melts, the extracellular medium becomes slightly more hypotonic, water goes inside the cells, the cell volume increases and the cell membrane breaks [10].

A basic cryotherapy protocol to be used in maxillary neoplastic lesions is based on repetitive cycles of freezing, slow defrosting, and maintenance of the frozen tissue for a period of 5 to 10 minutes, directly in the affected area, to help the removal of neoplasia or abnormal cell elements, without the need for extending the surgical margin. Liquid nitrogen is the most common and efficient option when cryotherapy is chosen for the treatment. In such case, immediate and long-term consequences of cryotherapy must be taken into consideration when choosing the treatment whenever important anatomic structures such as the airways are close to the area to be treated using this method.

Exacerbated local inflammatory reaction caused by cryotherapy might provoke severe edema within the hours following the procedure. In addition, hemorrhage during the surgery, immediately after the local defrosting process is a strong possibility. Also, although cryosurgery causes necrosis of the host bone cells, it keeps the inorganic bone structure, which, despite acting as the substrate for the formation of a new tissue, might result in mandible weakness and risk of pathological fracture [11].

This study aimed to present two clinical cases of odontogenic myxoma in adolescent patients. One of the patients was treated using a segment resection aided by a custom 3D-printed marginal bone debriding followed by liquid nitrogen cryotherapy.

Case Presentation

Case 1

A 14-year-old female patient was referred by another professional with a panoramic radiography showing osteolytic signals on the left mandible ramus. She presented absence of intra or extra oral signs or any other clinical symptom. The tomographic image revealed broad radiolucent lesion involving the left mandible ascending ramus, which was well delimited and with a multilocular aspect and septate by straight bony trabeculae (Figure 1A-1C). Curettage with incisional biopsy was carried out with access through the lesion vestibular face. The tissue presented a light non-gelatinous soft substance all over the exposed area (Figure 1D). The histopathological examination revealed conjunctive tissue with myxoid areas related to increased cell areas containing fusocellular elements.

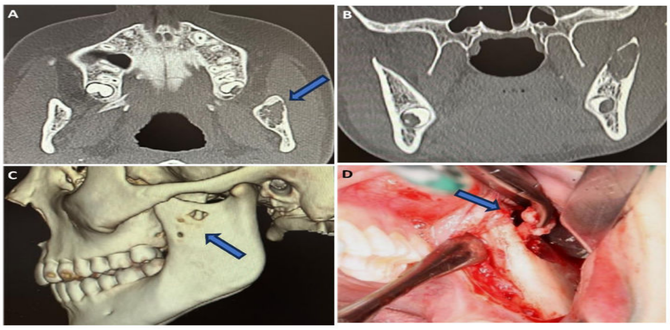

Figure 1. Preoperative computed tomography evidencing osteolytic lesion of the mandible ascending ramus. (A): Axial cross section; (B): Coronal cross section; (C): 3D virtual reconstruction; (D): Incisional biopsy.

The tomographic reconstruction studies determined the lesion limits and the virtual planning, for the construction of an osteotomy surgical guide that could guarantee an exeresis with suitable safety margins. By associating CAD-CAM and 3D tomographic imaging, a surgical 3D guide was prepared using a computer program (DDS-Pro-JST Sp. z o.o., Częstochowa, Poland) in a filament printer along with the mandible virtual biomodel, and the perfect adjustment of the parts was confirmed (Figure 2). The lesion extension allowed a design with preservation of structures such as the mandible foramen, lower alveolar nerve, and mandible condyle.

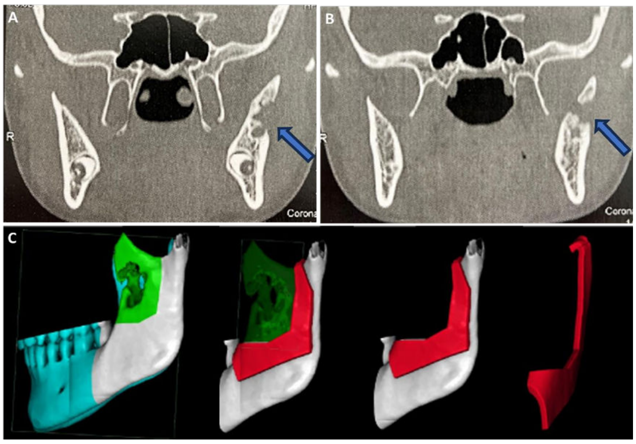

Figure 2. (A and B): Lesion tomographic images six months after the biopsy; (C): Virtually produced customized osteotomy 3D guide, based on the lesion limits and safety margin (DDS-Pro-JST Sp. z o.o., Częstochowa, Poland).

The surgical procedure was carried out under general anesthesia and nasotracheal intubation. The intraoral access was *via* vestibular incision to the left mandible ascending ramus, which allowed the visualization of the whole ramus and coronoid process. After adapting the 3D-printed osteotomy guide supported by the mandibular notch, the segment resection was performed aided by the piezoelectric device and complemented with multi-laminate drills up to the limit of the surgical guide (Figure 3).

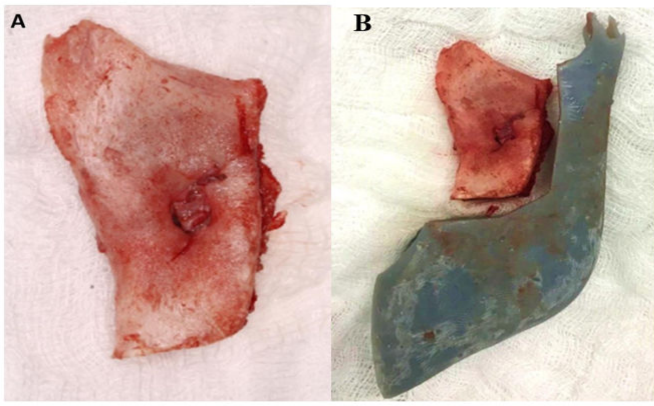

Figure 3. (A): Coronoid process resected from the mandible; (B): 3D biomodel of the resected mandible confirming the safety margin.

The histopathological examination of the surgical sample definitely confirmed the odontogenic myxoma diagnosis by the proliferation of fusiform cells in a star-like arrangement in myxoid stroma (Figure 4).

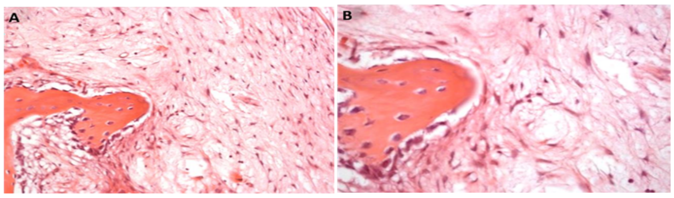

Figure 4. Histological optical microscopy confirmed the odontogenic myxoma diagnosis. Cross section of (A): 20x; (B): 40x.

In a four-year postoperative follow-up, good mouth opening and preserved mandibular movements were observed as well as absence of neuropraxic lesion or facial asymmetry. The tomographic examination presented a surgical area with normal bone neoformation and without evidence of lesion recurrence (Figure 5).

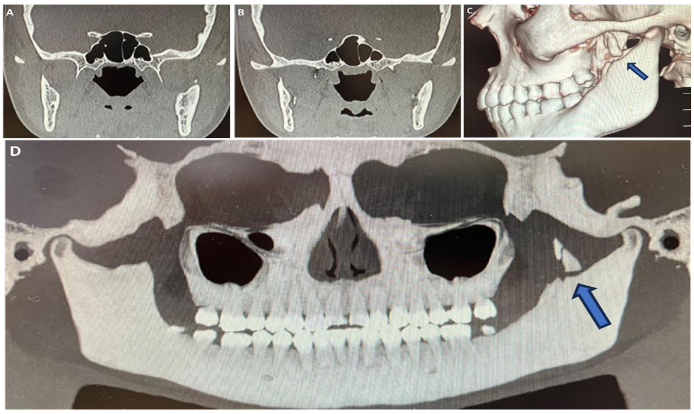

Figure 5. (A): Postoperative tomographic aspect after four years; (B): Showing normal bone repair; (C and D): With supposed formation of a new coronoid process.

Case 2

A 13-year-old male patient assisted by the cleft lip and palate referral service appeared in a routine appointment presenting a panoramic radiography where a radiolucent image was observed in the right mandibular body basal region, with teeth 47 and 48 involved in the lesion, which extended to the mandibular ramus (Figure 6A). In the clinical examination, he did not present any apparent symptom or signal. He was submitted to the tomographic examination, which confirmed a well-circumscribed osteolytic area, with well-defined limits and resorption of the surrounding cortical bone (Figure 6B).

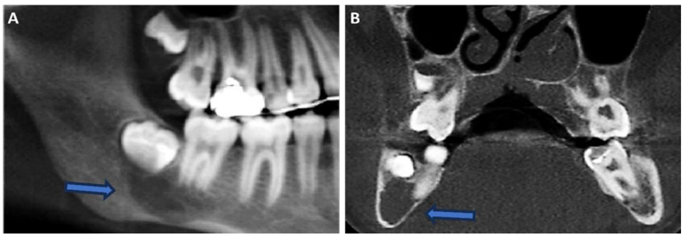

Figure 6. (A): Initial radiographic aspect; (B): Tomographic aspect showing a translucent central lesion on the right mandibular basal region.

The incisional biopsy was carried out through the vestibular access in the mandible angle and body, along with the simultaneous 48 extraction. The microscopic analysis revealed neoplastic areas of predominantly mesenchymal origin, composed by myxoid conjunctive tissue, with a mucoid matrix and areas of fibrous aspect (Figure 7). These characteristics associated with the tomographic examination allowed the conclusion of an odontogenic myxoma diagnosis.

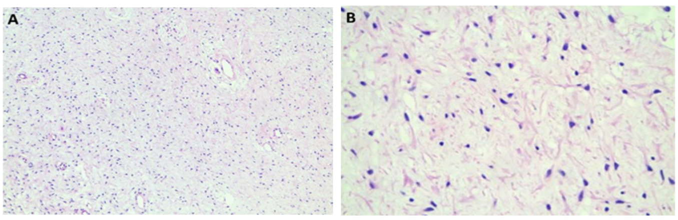

Figure 7. Histological optical microscopy confirmed the odontogenic myxoma diagnosis. Cross section of (A): 10x; (B): 40x.

After obtaining the diagnosis, the curettage of the lesion associated with peripheral osteotomy and followed by liquid nitrogen cryotherapy was planned. When the patient was prepared (serum exams and pre-anesthetic evaluation) the surgical procedure was carried out in hospital, under general anesthesia and nasotracheal intubation. The intraoral access included intra-sulcular incision vestibular to teeth 44, 45, 46, and 47, associated with an oblique anterior incision, vestibular to tooth 44. Tooth 47 was extracted due to its involvement in the lesion, and a surgical window was created by using multiblade burs to access the lesion and allow the curettage. The material obtained from the curettage (remaining bone fragments, and tooth 47) were all sent to the histopathological microscopic examination. After the curettage.

A peripheral osteotomy was performed using large diameter multiblade spherical burs for debriding the lesion margins and eliminate tumor remnants (Figure 8A and 8B). Dry gauze was interposed between the exposed bone tissue and the oral mucosa and around the whole tumor area aiming to protect the adjacent structures that would not be frozen. Next, the cryotherapy treatment started with liquid nitrogen spray in the tumor cavity. The applications on the cavity walls were carried out in three times of one minute in continuous spray with 5 minute intervals between each application, covering all the walls of the cavity and repeating the same protocol up to the conclusion of the applications (Figure 8C and 8D).

After the end of the suture and removal of the oropharyngeal cover, the patient presented profuse bleeding in the operated region. The suture was removed, and the flap reopened to identify the bleeding origin. It was observed that the hemorrhage originated in the lesion basal region and the floor of the mouth, close to the mylohyoid muscle. To control the bleeding and prevent future episodes, the areas were cauterized. The region was sutured again, and no bleeding was identified in the immediate and late postoperative period.

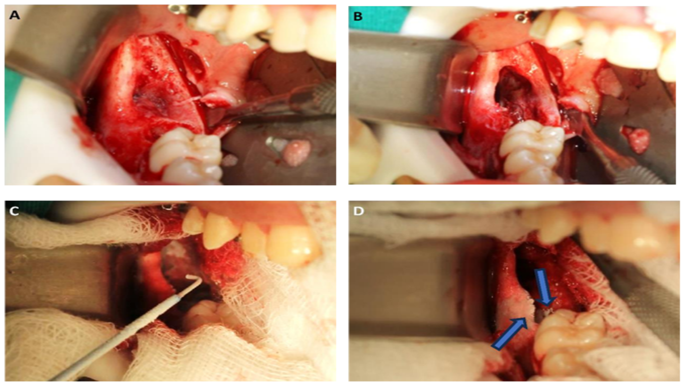

Figure 8. (A): Flap exposing the bone crest edge; (B): Curettage followed by peripheral osteotomy; (C): Liquid nitrogen applications on the cavity walls; (D): Frozen bone crystals indicated by arrows.

The decision to keep the patient in nasotracheal intubation, sedated and under surveillance at the Intensive Care Unit (ICU) was taken due to the risk of exacerbated edema and obstruction of airways. The patient presented a relevant edema condition on the right submandibular region before the end of the 24 hour postoperative period, which confirmed the correct conduct of keeping the airways *via* nasotracheal intubation and ICU assistance. The patient was treated with antibiotic medication of empirical choice, 500 mg tranexamic acid at each 12 hours, 500 mg hydrocortisone soon after the procedure, which was kept with a 100 mg dose at each 6 horas during the first 24 postoperative hours and ice pack applied to right submandibular region every 20 minutes for 48 hours, which was substituted with hot pack applied after that period. Such procedures allowed extubation on the third day after surgery and hospital discharge on the fourth day, with peripheral action painkiller prescription, if needed. The histological analysis confirmed the diagnosis of fibromyxoma (Figure 9).

Tomographic follow-up at six and twelve months after surgery were carried out in the first year, and after that period, once a year for five years, and no signs of relapse were observed (Figure 10).

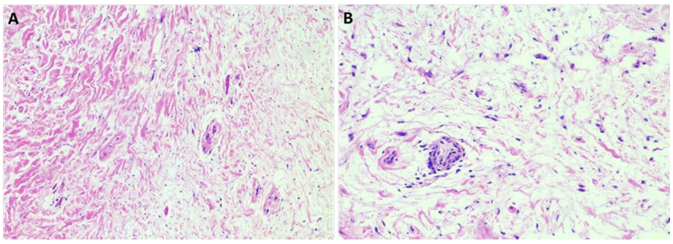

Figure 9. Histological optical microscopy confirmed the diagnosis of odontogenic fibromyxoma. Cross sections (A): 10x; (B): 20x.

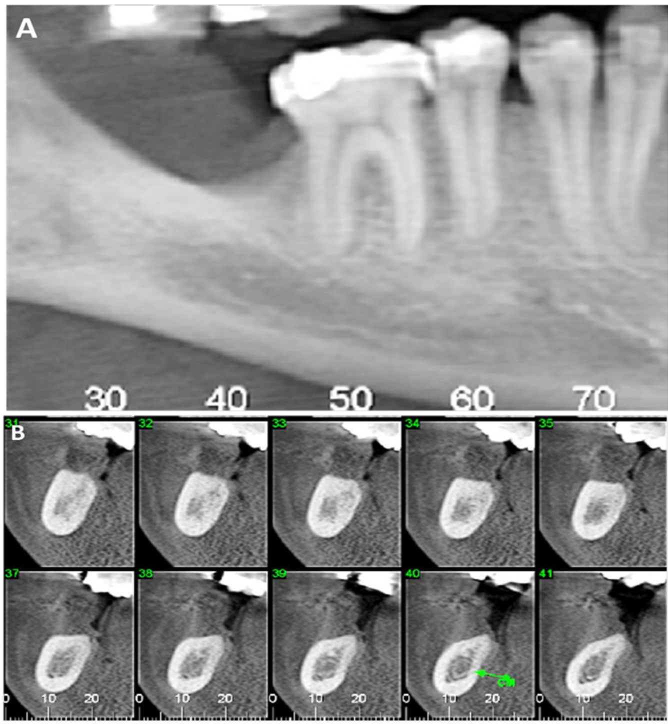

Figure 10. Tomographic evaluation after five years showing normal bone formation and no sign of relapse. (A): Panoramic cross section; (B): Coronal cross sections of the affected area.

Discussion

The odontogenic myxoma is a rare tumor and its diagnosis process demands effort in the search of clinical and imaging details. Its slow growth might lead to late diagnosis when it is already spread and has invaded adjacent anatomical structures. The myxoma treatment is unavoidably surgical, with several protocols already described in the literature and the technique of choice depends on its size and proximity of adjacent dental and neural structures [12].

In the first case reported, the guided surgery allowed the definition of a safety margin and the exact replication of the conduct planned, preserving neural structures and the mandibular condyle. The good evolution of that case is due to the lesion location, which although presenting moderate size, kept those structures out of the safety margin. In the 4-year follow-up, tomographic examinations showed a tumor free healing and bone growth in the resection foreground, which suggests new formation of the mandible coronoid process. Each case reported was diagnosed in individuals of different gender and adolescents. However, some authors have stated that OM shows no gender predilection, some studies have reported higher incidence in female and young patients [13].

In a review of 6,000 bone tumors in the Mayo Clinic, all myxoma cases reported were mandibular with a few cases affecting non-toothed areas [14]. The first case reported was diagnosed in an unusual location involving the mandibular ramus and part of the coronoid process, withouti nvolving toothed areas which, associated with the early diagnosis, allowed resection with no further damage.

Bone reconstructions using preoperative virtual models were already described for mandibular reconstruction [15]. Although the real time navigated surgical procedure can be used instead of printed biomodels, those computer softwares are complex and biomedical engineers must be involved. It seems to be relevant to emphasize that the evolution of stereolithographic models has improved the surgical predictability from the recreation of defect. And the production of resection guides.

Although the safety margin definition depends on the operator analysis during the virtual planning, the expectation is to achieve a quite accurate surgical procedure in relation to the design. In the case reported, the fact that it was a procedure carried out *via* intraoral access in a region of difficult visualization and surgical access, the use of a guide was an important help in the osteotomy reproduction.

In a follow-up of 13 cases of OM treated only with conservative approaches and a literature review, the authors observed 26% (2 cases) of recurrence. This rate, is similar to the recurrence results (25%) obtained after radical treatments, but radical treatments may be unavoidable for larger lesions, and may represent negative aesthetic and functional results that can decrease patients' quality of life [16].

The ideal management of benign lesions that present aggressive biological behavior and might relapse would be the one that provided, in addition to the full lesion exeresis the least functional and aesthetic impairment. Thus, cryosurgery outstands among the maxillofacial surgery. Although the surgical intervention is the treatment of choice for odontogenic myxomas, some issues have been raised in relation to the type of treatment to be used in each case. It seems to be relevant to emphasize that once the postoperative control is duly performed, an early diagnosed recurrent lesion does not imply into treatment failure and can be removed with a relatively simple procedure, without having to undergo major surgical interventions with difficult recovery [17].

We observed that the curettage with peripheral osteotomy of the lesion associated with the cryotherapy action mechanism as an ancillary procedure played a fundamental role in the relapse prevention in a five-year period. One of the cryotherapy choice criteria was the fact that it was a potentially relapsing tumor, in which case the resection with safety margin would be indicated, but probably leading to a functional and aesthetic loss of the jaw of an adolescent male (Case 2). However, some studies have reported that even in local relapse conditions without complications, repeating the cryosurgical approach might present viable outcomes, in addition to avoiding major bone defects and functional impairment.

Due to the approach in the mandibular body and angle region followed by a hemorrhagic condition, we opted for keeping the patient intubated, sedated and observed, since we predicted exacerbated edema of retromandibular and submandibular spaces due to the cryotherapy. This procedure prevented postoperative complications resulting from the impairment of airways.

The low temperatures applied reduce the bone repair capacity, which can extent up to 1 cm deep depending on the freezing method and local conditions [18]. Clinical studies already reported this situation from the fifth weeks after surgery onwards up to the twenty-fourth week, with an approximate mean of twelve weeks, which confirms experimental studies in which the cooled mandible showed severe weakening around the eighth week, mainly in lesions whose diameter was over 4 cm, becoming susceptible to pathological fracture episodes. Considering the characteristics of the bone defect caused in the case reported, the patient was instructed to keep a soft diet for fourteen weeks due to the inevitable frailty.

The application protocols used in the cryotherapy in maxillomandibular bone lesions range between 10 seconds and 8 minutes [19]. The former was the shortest time analyzed in a study using rabbits. However, several authors, in cases reports and literature reviews, follow basic principles of cryotherapy as an ancillary treatment of maxillomandibular bone lesions, by applying at least 2 cycles with spontaneous reheating intervals of at least 5 minutes [20]. In the second case reported in this study, the application of liquid nitrogen spray in three cycles of one minute each, with a five-minute interval for reheating between them, was performed. The procedure included all the walls of the cavity of a lesion diagnosed as odontogenic myxoma in mandibular body. The 5-year follow-up showed that the treatment was successful, without fractures, good healing evolution and no evidence of lesion relapse.

However, the absence of a defined therapeutic protocol allied to the lack of scientific method, since most of the studies are descriptions of the authors' clinical and surgical experiences, should encourage standardized research to securely state the effectiveness of the treatment.

Conclusion

The printed surgical guide, designed based on 3D virtual simulation, allowed a procedure with relatively short surgical time and easy resection *via* intraoral access, thus, justifying a more conservative surgical approach in the treatment of odontogenic myxoma of the mandible ramus. However, for the treatment of the same lesion in mandible body, the surgery associating curettage and debriding with liquid nitrogen spray cryotherapy applied with safety procedures allowed a quite conservative treatment, without bone resection and minimum consequences, resulting in the absence of radiologic signals of relapse for 5 years.

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