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Osteoma in the temporomandibular joint (Case report)
Osteoma en la articulación temporomandibular (Caso clínico)

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Abstract:

Osteomas are benign slow growing osteogenic tumors mostly arising in the craniofacial region and characterized by the deposition of differentiated and mature either or both cancellous and compact bone. Osteoma accounts for 2-3% of all bone primary tumors with an incidence of 10-12% among all benign skeletal neoplasms. **Objective:** The objective of this work is to describe a clinical case of an osteoma in the temporomandibular joint diagnosed in the maxillofacial surgery service of the General Hospital of Pachuca in the State of Hidalgo, Mexico. **Clinical Case:** A 39-year-old female patient who comes to the General Hospital Pachuca, Mexico due to pain and noises in the right preauricular region of 6 years of evolution, with facial asymmetry, mandibular deviation to the left and limited mouth opening. Clinically, facial asymmetry, mandibular deviation to the left, right posterior crossbite, anterior open bite mainly on the affected side, preauricular pain, joint sounds, and limitation of mandibular movements were observed. Radiographic examination revealed a trapezoidal mass measuring 2.5 by 2.0 cm, with alteration of the condyle-mandible anatomy on the right side. An insertional biopsy is performed, reporting an osteoma, and surgical intervention is continued. **Conclusion:** The osteoma in the temporomandibular joint is a rare lesion, its timely value is essential for its treatment. Surgical resection is the gold standard treatment, which is based on a radical excision that extends to the altered normal bone, with the contextual objective of achieving an optimal aesthetic result by choosing the least invasive surgical treatment possible.

Keywords:

Osteoma, Temporomandibular Joint, Jaw

Resumen:

Los osteomas son tumores osteogénicos benignos de crecimiento lento que surgen principalmente en la región craneofacial y se caracterizan por el depósito de hueso esponjoso y compacto diferenciado y maduro. El osteoma representa el 2-3% de todos los tumores primarios óseos con una incidencia del 10-12% entre todas las neoplasias esqueléticas benignas. **Objetivo:** El objetivo de este trabajo es describir un caso clínico de un osteoma en la articulación temporomandibular diagnosticado en el servicio de cirugía maxilofacial del Hospital General de Pachuca en el Estado de Hidalgo, México. **Caso Clínico:** Paciente femenino de 39 años que acude al Hospital General Pachuca, México por dolor y ruidos en la región preauricular derecha de 6 años de evolución, con asimetría facial, desviación mandibular hacia la izquierda y limitación a la apertura bucal. Clínicamente se observó asimetría facial, desviación mandibular hacia la izquierda, mordida cruzada posterior derecha, mordida abierta anterior mayormente del lado afectado, dolor preauricular, ruidos articulares y limitación de los movimientos mandibulares. Al examen radiográfico se observa una masa de 2.5 por 2.0 cm, de forma trapezoidal, con alteración de la anatomía cóndilo-mandíbula del lado derecho. Se realiza biopsia insercional reportando osteoma, se prosigue a realizar intervención quirúrgica. **Conclusión:** El osteoma en la articulación temporomandibular es una lesión poco frecuente, su valoración oportuna es fundamental para su tratamiento. La resección quirúrgica es el tratamiento estándar de oro, la cual se basa en una escisión radical que se extiende hasta el hueso normal circundante, con el objetivo contextual de lograr un resultado estético óptimo mediante la elección del tratamiento quirúrgico lo menos invasivo posible.

Palabras Clave:

Osteoma, Articulación temporomandibular, Mandíbula

INTRODUCTION

Osteomas are benign slow growing osteogenic tumors mostly arising in the craniofacial region and characterized by the deposition of differentiated and mature either or both cancellous and compact bone.[1]

There are three types: peripheral, central or extraskeletal.[2] Peripheral type arises from the periosteum, central type arises from the endosteum and extraskeletal type arises within the

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muscles or dermis.[1] The exact etiology of osteomas is unknown and is multifactorial.[2] It may be genetic (as seen in Gardener's syndrome), related to endocrine disorders, trauma or even inflammation.[3]

Osteoma accounts for 2-3% of all bone primary tumors with an incidence of 10-12% among all benign skeletal neoplasms.[1,2] It is more frequent in adolescents and young adults with 50% of patients being aged between 10 and 20 years and it predominates in male, with a reported male-to-female ratio of 4:1.[4]

The age range for presentation of this lesion is very wide with the average age of diagnosis as 50 years.[1] Among the jaws, mandible is more commonly affected than maxilla with angle and condyle being the most common site followed by the body (molar region) and ascending ramus.[5] Interestingly these are also the sites for various muscle attachments on the mandible.[1]

Jaw osteomas are usually asymptomatic except for some large lesions that may cause functional disturbances or neurovascular compression symptoms due to their large size.[2,3]

Condylar osteomas may cause reduced mouth opening, occlusal disharmony, facial asymmetry and even in the temporomandibular joint dysfunction.[6] Most peripheral osteomas present within the paranasal sinuses are usually associated with sinus infections while some even develop within a sinus polyp, a finding which does not rule out the role of infection in the development of an osteoma.[3]

Most central lesions have been reported in the premolar–molar region of the mandible.[2] Central osteomas arise within the endosteum and hence cause expansion of one or both the cortices.[1]

Root displacement can also be seen if the lesion involves a dentulous area.[7] Some tumors are associated with pain and neurologic disturbances when compression of adjacent nerves is associated with the lesion due to its position and growth.[5]

CLASSIFICATION

Depending on the site, three subtypes of osteomas are distinguished:

- Central: characterized by progressive endosseous development, eventually resulting in the complete replacement of the affected bone segment.[3]
- Peripheral: consisting of periosteal development that can appear as a pedunculated mass.[3]
- Extraosseous: which develops within the soft tissues, particularly in the muscles.[1,3]

PATHOGENESIS

The pathogenesis of osteomas is still under debate, and different onset sites are described in the literature, such as the frontoethmoidal junction or the temporal bone, where osteomas can be associated with congenital cholesteatoma.[7] For this reason, some authors consider osteomas to be congenital lesions, arising from an embryonal cartilaginous rest or a persistent embryological periosteum.[8] The association of osteomas with colonic diseases such as Gardner's syndrome suggests a possible hereditary nature.[9] On the other hand, some of the most frequent osteoma onset locations are susceptible to trauma (e.g., to the frontal bone or to the angle and lower border of the mandible), suggesting that previous trauma may contribute to the development of these tumors.[1] Osteomas exhibit continuous growth rather than growth cessation.[4]

ETIOLOGY

The exact etiology of osteomas is unknown and is multifactorial.[1] It may be genetic, related to endocrine disorders, trauma or even inflammation.[10] The neoplastic nature of osteomas is doubted due to their limited growth potential and absence of recurrence.[2] A combination of trauma and subsequent muscle traction may play a role in the development of osteomas as trauma causes subperiosteal bleeding that locally elevates the periosteum.[4] The continuous muscle traction accentuates the osteogenic reaction stimulated by local bleeding.[6]

Although the patient in this case had a history of direct trauma prior to development of the lesion, this theory of combination of local bleeding and muscle traction does not seem to be true for this case as this is an endosteal osteoma and only the development of periosteal osteoma can be influenced by muscle traction.[1] Another etiological theory based on stimulation of embryological remains states that the membranous and cartilaginous elements present at the sutures of skull bones could stimulate the development of cell rests and subsequent tumor formation.[8]

It could be hypothesized that it is the stimulation of osteogenic cells (differentiated or embryonic cell rests) by any means like infection, trauma, genetic alteration which could lead to the development of an osteoma.[11]

CLINIC

Clinical presentation can vary depending on the location of the lesion; patients generally complain of localized, deep, and unremitting pain that increases in intensity over time, typically gets worse at night and rapidly improves after salicylates or other NSAIDs administration.[2] Soft tissue swelling and skin erythema may be present in osteomas in subcutaneous location.[5] Duration of pain can vary from weeks to years before definitive diagnosis, depending on how typical the clinical presentation is and how early the diagnosis suspect is posed.[8]

Osteomas can assume significant dimensions causing aesthetic and/or functional problems due to bone distortion, with possible compression of nearby structures.[1] Clinical manifestations of craniofacial osteomas are highly variable according to the sites of onset, as suggested by multiple case reports described in the literature.[12] The most frequent osteoma onset sites are the jaw and the paranasal sinuses (frontal, ethmoid, maxillary, and

sphenoid), followed by the internal and external cranial planking and the maxillary bone.[6]

DIAGNOSIS

The most used images for its diagnosis are panoramic radiography and computed tomography, in addition to bone scintigraphy to confirm whether it is in active or inactive growth.[3] The diagnosis is often made incidentally through radiological investigations conducted for other reasons.[5]

On radiographic assessment the lesion appears as 'bone within bone' with both the cortices expanded and intact with the medulla of the lesion confluent with the adjacent normal bone.[8] X-ray is the initial imaging modality in case osteoma is suspected.[13] Plain radiograph typically shows a round or oval radiolucent nidus, usually smaller than 1,5 cm in diameter, surrounded by a variable but regular fusiform area of bone sclerosis.[8] In larger lesions, a central nucleus of bone mineralization within the nidus can also be observed.[10] Sometimes the peripheral rim of reactive bone sclerosis is so extensive that the underlying radiolucent nidus may result obscured.[11]

Computed Tomography is the imaging technique of choice for osteoma, being advantageous for detection and characterization of both the nidus and the peripheral sclerosis.[3] It is particularly helpful for small osteomas, for those lesions which show less peripheral bone sclerosis (especially medullary and subperiosteal types), and in all those cases where standard radiographs are not clearly conclusive.[14] On CT scan the nidus appears as a small well-defined radiolucent area, with soft-tissue attenuation values, which shows early vascular contrast enhancement.[15]

HISTOLOGICAL FINDINGS

Histologically, an osteoma is represented as a mass of abnormal dense bone that may originate from the periosteum or from bone marrow, thus differentiating between two types of osteomas.[1] Compact osteomas, also called "ivory", are made of mature lamellar bone with minimal marrow spaces and occasional haversian canals without any fibrous structure.[4] On the other hand, trabecular osteomas, also called "mature" are composed of cancellous trabecular bone with bone marrow enclosed by a cortical bone margin.[6] This distinction is typical of all osteomas and does not involve any association with a pattern of tumor proliferation.[1]

DIFFERENTIAL DIAGNOSIS

Differential diagnosis of osteoma includes complex odontoma, central ossifying fibroma, osteoid osteoma, osteblastoma, Pindborg's tumor and low-grade osteosarcoma.[3]

Odontomas are hamartomas of hard tissue of dental origin.[1] The radiographic appearance of odontomes varies from mixed to radiopaque depending on its developmental stage.[4] Compound odontomes appear as well-defined irregular tooth like radiopacities of varying sizes with a surrounding radiolucent band.[2] They may be single or multiple in number and rarely exceed 30 mm in size, thus making only complex odontoma as a possible differential diagnosis.[9]

Central ossifying fibroma, earlier termed as 'cementifying ossifying fibroma' is a benign fibroosseous lesion.[8] It has a peak incidence at around 20–40 years of age with the lesion mostly occurring in mandible and affecting females more than males.[6] Radiographically they appear as unilocular or multilocular radiolucencies having well defined corticated borders with or without multiple radiopaque foci or flecks of varying radiodensities and sizes depending upon its maturity.[16] When totally calcified, it may appear as completely radiopaque.[3] With sufficient growth they may cause cortical expansion, tooth displacement and even resorption.[5]

Osteoid osteoma is a benign osteogenic neoplasm with limited growth potential.[17] Mandibular cases have been reported more than those in maxilla.[16] Radiographically, it presents as a well-defined round to oval radiolucency called the 'nidus' with varying amount of radiopacity in the centre and peripheral reactive sclerosis.[15] It rarely exceeds 2 cm in size.[1] Nocturnal pain which is alleviated by NSAIDs is the most striking feature of this lesion.[17] Cortical and subperiosteal osteoid osteoma do not cause expansion while cancellous or medullary variant may cause an obvious cortical expansion.[16]

Osteoblastoma is also a benign osteogenic neoplasm mostly affecting the axial skeleton in young adults.[15] Among the jaws mandible is the more common site.[18] It differs from osteoid osteoma in its higher growth potential with most lesions exceedingly more than 2 cm in size.[13] They cause pain which is not relieved by NSAIDs along with local swelling and tenderness.[16] Radiographic presentation comprises of well-defined radiolucency with areas of patchy radiopacities of varying densities and peripheral sclerosis.[18] A radiolucent band is usually visible in a mature lesion.[17] The main distinguishing features of Central ossifying fibroma from central osteoma is its surrounding radiolucent band and its well delineated and corticated borders.[15]

Calcifying epithelial odontogenic tumor is an uncommon benign tumor of odontogenic origin mostly occurring in 20–40 years of age without any sex predilection and occasionally associated with an unerupted tooth.[18] Most cases have been found in the posterior mandible.[14] Cortical bone perforation and tooth displacement are common findings in the central variant.[17] Radiographic appearance varies from completely radiolucent to mixed radiopaque and unilocular to multilocular depending on its developmental stage.[18] The margins are usually well defined.[12] Histologically, the tumor comprises of polyhedral epithelial cells arranged in sheets in a fibrous background, Liesegang's rings and variable amount of amyloid like material.[16] Osteosarcoma is a primary malignant tumor of cells of mesenchymal origin having ability of osteogenic differentiation.[17] Clinical features include fast expansile growth with pain, paresthesia, and tooth mobility.[14] Radiographic

appearance may be of a completely radiolucent, mixed, or radiopaque lesion with ill-defined borders with occasional root resorption and cortical breach due to its aggressive nature.[18]

TREATMENT

Surgical resection is the gold standard treatment.[1] It is based on a radical excision extending to the surrounding normal bone, with the contextual aim of achieving an optimal cosmetic result by choosing the most minimally invasive surgical treatment possible.[16] Over decades complete surgical excision has been considered the classical treatment for all those patients who had unremitting pain despite conservative treatment or those who could not tolerate long-term NSAIDs therapy.[4]

En bloc surgical resection has a reported success rate between 88% and 100% but it carries several significant disadvantages.[19] Firstly, for the orthopedic surgeon it may be difficult to identify the precise location of the lesion and the exact amount of bone to resect; secondly, to be sure all the nidus is removed, a substantial volume of bone must be excised, potentially leading to bone weakness and consequent necessity of bone grafts, internal fixation, and postoperative immobilization to prevent subsequent fractures.[18] Nonetheless, recurrence rate remains significantly high (reported in literature to range from 4,5% to 25%) due to incomplete nidus excision.[20]

OBJECTIVE

The objective of this manuscript is to describe a clinical case of an osteoma diagnosed in the maxillofacial surgery service of the General Hospital of Pachuca in the State of Hidalgo, Mexico.

PRESENTATION OF THE CLINICAL CASE

A 39-year-old female patient who comes to the General Hospital of Pachuca, Mexico due to pain and noises in the right preauricular region: the anamnesis shows that this condition began when she was 33 years old with approximately 6 years of evolution. Clinical examination of the head revealed facial asymmetry, mandibular deviation to the left, right posterior crossbite, anterior open bite mainly on the affected side, preauricular pain, joint sounds, and limitation of mandibular movements.

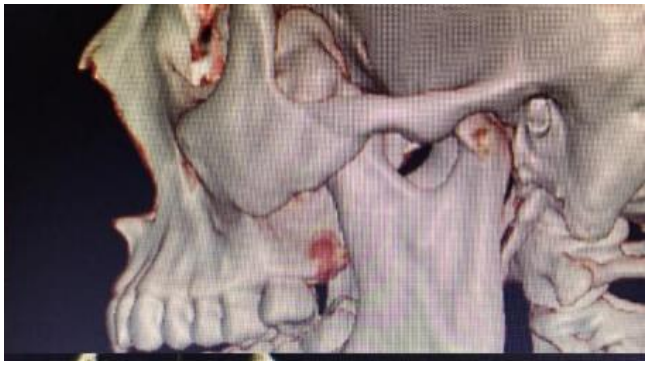

Figure 1: 3D image of a lateral skull showing an osteoma in the temporomandibular joint.

Extraoral clinical examination revealed a deformation in the right preauricular region due to the presence of tumor masses in the region of the condyle of the right mandible: fixed, multilobulated, hard lesions were evident on palpation. Computed tomography studies and 3D images of the affected facial mass were performed, observing a trapezoidal mass of 2.5 by 2.0 cm, with alteration of the condyle-mandibular anatomy on the right side (Figures 1-3). With clinical and radiographic data, it was determined that it was an osteoma in the region of the temporomandibular joint. Surgical intervention continues.

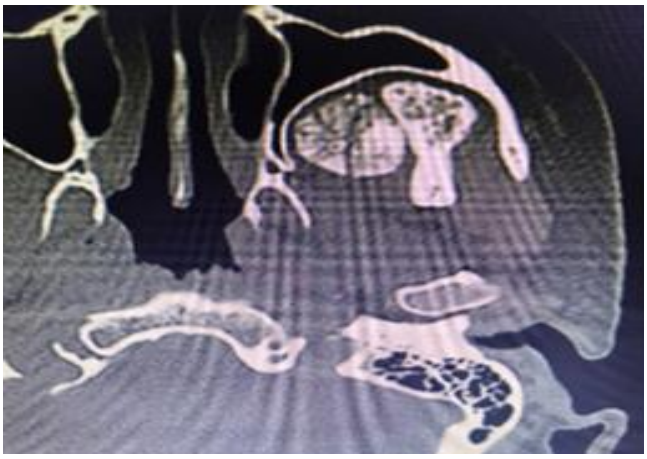

Figure 2: Computed tomography of the base of the skull where an increase in volume is observed in the temporomandibular joint.

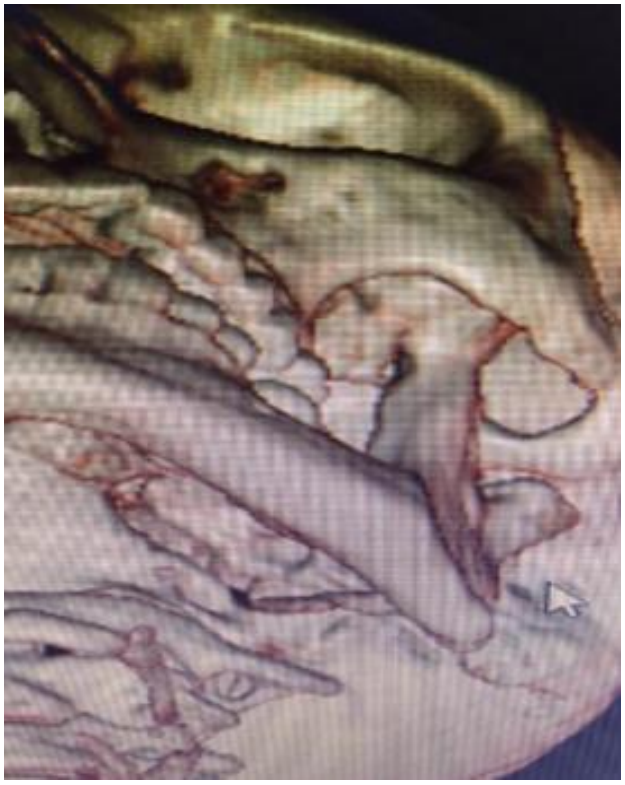

Figure 3: 3D image of the skull where an increase in volume is observed in the temporomandibular joint.

DESCRIPTION OF THE TECHNIQUE

Under balanced general anesthesia, nasotracheal intubation, infiltration of 2% lidocaine with epinephrine 1:100,000 for hemostatic purposes, through a preauricular approach, it is dissected by plans, vascular structures are identified, until the bone surface of the temporomandibular joint and tumor are exposed. Marginal osteotomies are performed, which was eliminated by means of a lower right condylectomy with a 1-mm margin of healthy tissue with the help of an electric-type pneumatic saw, chisel and percussors (Figure 4). Once the tumor mass has been removed, residual remodeling of the compromised region is carried out with low-speed rotary instruments and constant irrigation with physiological solution. Secretions are aspirated, drenovac 1/8 is placed, it is sutured in planes with vicryl 3-0 and skin with nylon 4-0 and the surgical procedure is terminated without incidents or immediate trans or postoperative accidents, the patient leaves partially recovered from the operating room.

The removed surgical specimen was sent for a pathological study for histopathological diagnosis, which reported the presence of dense cortical bone with a lamellar pattern and scant medullary spaces, concluding with the diagnosis of osteoma (Figure 5). The patient remains hospitalized for 5 days after surgery for antibiotic impregnation, after a month a tomography of the facial mass is requested.

ETHICAL CONSIDERATIONS

The manuscript was written in accordance with the precepts of the Declaration of Helsinki and informed consent was obtained from the patient to publish the images (always respecting confidentiality and concealing the patient's identity).

DISCUSSION

Osteoma is a benign tumor characterized by a variable amount of calcification, sclerosis and bone, the growth of these tumors is limited, with lesion size generally less than 2 cm according to the WHO classification criteria.[15]

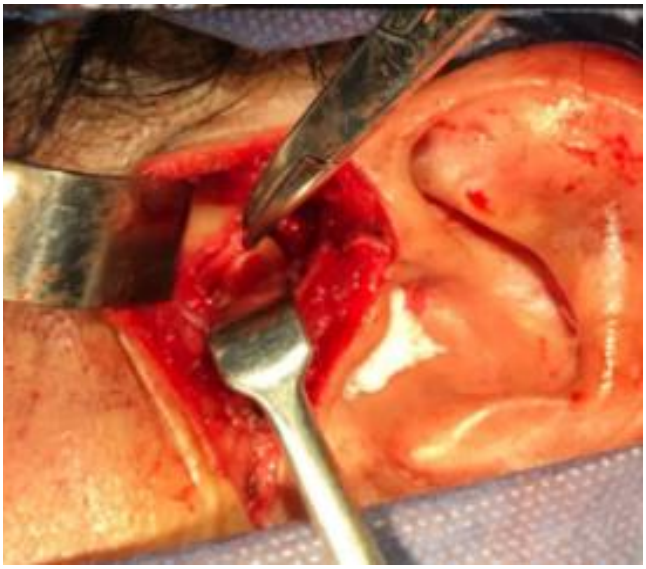

Figure 4: Surgical procedure showing surgical dissection by planes of the temporomandibular joint.

Osteoma is extremely rare in the head and neck region.[16] About 80% of osteomas occur in the long bones, while less than 1% occur in the faces.[17] Its actual incidence and prevalence are unknown since most cases are asymptomatic.[18] It is the most common benign neoplasm of the paranasal sinuses and the orbit, and can present at any age, in most cases, between the 2nd and 5th decades of life.[19] In the craniomaxillofacial region, most of these tumors occur in the mandible instead of the maxilla, causing spontaneous pain, and most are treated with excision of the damaged tissue.[20] They usually present as incidental radiological findings.[21] They are normally solitary masses and when they occur multiple times, we must rule out the presence of a syndromic complex such as Gardner's syndrome.[22] Peripheral osteomas of the mandible are uncommon, being more frequently located in the vestibular and basal cortices.[23] In our clinical case, the osteoma was found in the condyle of the right mandible, which presented symptomatically, since its growth exceeded the limits of the bone where it sits, causing an asymmetry or facial deformity. The individualized evaluation of

the size and location of the tumor was a starting point for planning the treatment and its complications derived from his surgery. The literature recommends an annual radiographic control of osteomas, especially those that cannot be completely removed in surgery, such as those of the orbit and sphenoid sinus.[24] As for the symptomatic mandibular osteomas, as in our case, they were removed with the appropriate intraoral approach since the patient already had an increase in size in the preauricular area that affected her daily life.[25] Having established surgical treatment as the treatment, an adequate approach was planned based on the size and location of the tumor, and the skill of the surgical team, knowing that recurrence is possible, although rare.[1] That is why it is important to create an adequate field of vision to avoid surgical complications.[2] It should be borne in mind that osteomas grow from the center to the periphery, so their partial resection, leaving a peripheral remnant, will very rarely give recurrence and, therefore, their complete resection in critical areas will not be necessary when the risk of surgical damage is high.[12] The differential diagnosis of osteoma is osteoblastoma.[26] Both osteoblastoma and osteoma are benign osteoblastic lesions of bone.[13] They have similar histologic features and likely represent different expressions of one pathologic process.[4] Osteoma is small, self-limited, and benign, whereas osteoblastoma is often larger (>2 cm), more aggressive, and may become malignant .[27] Osteomawas distinguished from osteoblastoma in the present case based on clinical features, radiographic features, and histologic.

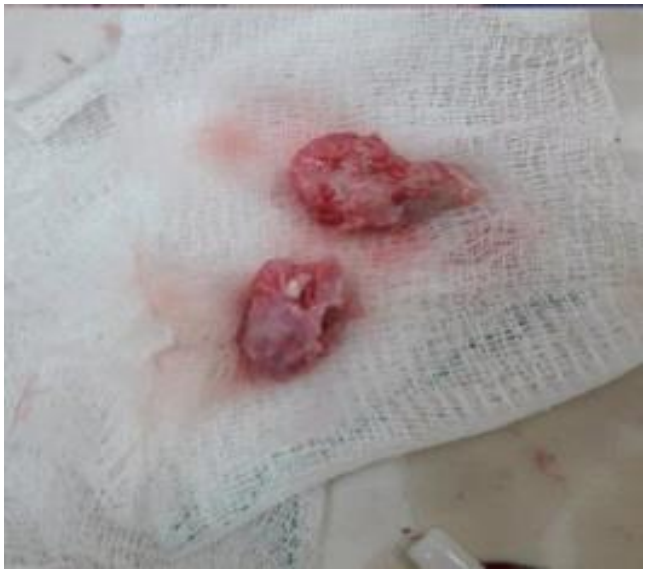

Figure 5: Osteoma of 2.0 x 2.5 cm removed by condylectomy and fragmented into 2 pieces.

CONCLUSION

Osteoma in the temporomandibular joint is a rare lesion, usually at the time of diagnosis it is accompanied by alterations that prevent the normal performance of the individual due to the discomfort it causes. The correct diagnosis and treatment are of vital importance, because this ensures a good prognosis of the disease.

FINANCING

None.

INTEREST CONFLICT

The authors declare no conflict of interest.

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