

Osteoma of maxillary sinus: case report

Osteoma del seno mascellare: descrizione di un caso clinico

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Parole chiave

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Summary

Cranio-facial osteomas are frequent in the nasal and paranasal sinuses, particularly in fronto-ethmoidal sites; other sinus cavities are more rarely affected. Although various theories (embryogenetic, traumatic and inflammatory) have been advanced to explain the pathogenesis, it is difficult to establish a specific cause-effect relationship. Nasal and paranasal osteomas are generally asymptomatic and are diagnosed on the basis of X-rays performed for other conditions, the onset of sinusitis-like symptoms or the appearance of complications due to sinus diseases. These cases require surgical removal to avoid the risk of short- or long-term complications or to solve any that may already exist. Herein, a rare case of osteoma of the maxillary sinus is described and the possible aetiopathogenetic role of traumatic and inflammatory factors described. The main clinical and therapeutic data concerning this lesion are examined.

Riassunto

Gli osteomi del distretto cranio-facciale si localizzano frequentemente a livello dei seni naso-paranasali ed in particolare in sede etmoido-frontale. Più rare sono le localizzazioni nelle restanti cavità sinusali. Sebbene diverse teorie (embriogenetica, traumatica e flogistica) tentino di spiegarne la genesi, uno specifico rapporto di causa-effetto è difficilmente dimostrabile. Gli osteomi del distretto naso-paranasale sono generalmente asintomatici e vengono diagnosticati in corso di indagini radiografiche eseguite per altre patologie, per l'insorgenza di una sintomatologia simil-sinusitica o per la comparsa di complicanze. In questi casi si rende necessario un trattamento chirurgico di exeresi per preservare il paziente dal rischio di complicanze sia precoci che tardive o per risolvere quelle già presenti. Gli Autori descrivono un raro caso di osteoma del seno mascellare e discutono il probabile ruolo eziopatogenetico svolto da fattori traumatici e flogistici, inoltre riportano alcune considerazioni sui principali dati clinico-terapeutici di tale patologia.

Introduction

Osteomas are a type of benign bone-forming tumour occurring most commonly in the cranio-facial skeletal structures, mainly in the nasal and paranasal regions, presenting, most frequently, in young subjects in the second and third decade of life, with a male/female ratio of about 2/1¹.

In 95% of cases, the osteoma is situated in the fronto-ethmoidal region, involving the frontal sinus in 60-70% and the ethmoid in 20-30%, while it is only rarely found in the nasal cavities, with the exception of the sphenoid sinus²⁻⁴.

In the upper maxillary region (5% of cases), the osteoma may appear in the form of a limited peripheral lesion involving the alveoli or cheek or as a tumoural growth developing inwards towards the sinus.

Although from an aetiological viewpoint, these lesions have been correlated to abnormal enlargements of embryonal tissues, previous cranio-facial traumas

(20% of cases) or chronic inflammatory processes of the nasal and sinus structures, a specific cause-effect relationship between osteomas and the triggering event still remains to be defined^{1,2,5}.

Nasal and paranasal osteomas are usually asymptomatic and are almost always diagnosed in the course of X-ray examination for other disorders. However, in spite of their slow evolution, it is not rare for such osteomas, particularly the fronto-ethmoidal type, to develop ophthalmological and meningo-encephalic infections⁶. In fact, as the mass increases in size, it may occlude the sinus ostium and thus block mucociliary activity leading to retention of secretion and the onset of sinusitis or mucocele.

Surgical treatment is reserved for patients with rapidly growing osteomas or infection and/or complications due to compression associated with severely painful symptoms, in the head and face^{1-3,7}. In the case of large or solid osteomas situated at maxillary sinus level, the incision can be external or a combination of intranasal and sublabial. The approach is

the median wall of the maxillary sinus. At the final histological examination, the material removed was found to be compatible with an osteoid osteoma (Fig. 4). CT scan of the mid-face bone structures performed 2 months after surgery showed no signs of relapse (Fig. 5) and the last examination, after 4 months, showed the patient to be free from signs and symptoms of disease recurrence.



Fig. 4. Osteoid osteoma: histological examination showed wide trabeculae of mature bone, lipomatous bone marrow and decalcification artifact.



Fig. 5. CT scan performed 2 months after surgery.

Discussion and conclusions

Osteomas of the maxillary sinus are benign tumours of slow evolution. In fact, they represent only 5% of the tumours involving this site. The pathogenesis of these tumours still remains to be elucidated, but several theories have been advanced in this respect.

The theory regarding embryological causes suggests that these osteomas arise from osseous proliferation due to the apposition of membranous and enchondral tissue⁸ forming particularly close to the bone sutures, such as the fronto-ethmoidal one. However, this would not explain why osteomas occur also in other parts of the body. Since a skeletal trauma gives rise to a proliferative remanagement of the bone, the theory of a traumatic origin has been suggested in a number of papers, according to which the development of these tumours is due to such events in the past (in 20% of cases), particularly in males and during puberty, when skeletal growth is at its peak^{2,8,9}. This theory, however, fails to explain why some osteomas occur in older patients or in those with no history of previous traumas.

Other theories suggest a determinant role may possibly be played by a previous inflammation of the sinuses, which would act as a stimulus for the proliferation of osteoblasts situated along the muco-periosteal join of the paranasal sinus; the calcification surrounded by inflamed tissue would subsequently give rise to the osteoma. However, some osteomas do not coincide with this theory.

The two latter theories, suggesting traumatic and inflammatory causes, have often been claimed to be responsible for osteomas of the maxillary sinus, on account of the particular anatomical position of this sinus, which is often exposed to recurrent traumas and tends to easily present infections and inflammatory conditions. However, the fact that osteomas occur only rarely in this site would seem to exclude the hypothesis of a casual role by these factors. In addition, the characteristic histological structure of spongy and/or eburneous osteomas would not reflect a hyperplastic bone reaction from trauma or infection⁹⁻¹². It is also difficult to assess the cause-effect relationship over a long period of latency of the clinical symptoms, which is why the pathophysiology of these lesions is still unknown^{1,2,5,10}.

However, the fact that our patient had undergone a Caldwell-Luc operation for chronic sinusitis, nine years previously, strongly supports the hypothesis of this causal relationship, in this case. The irritative stimulus of the chronic inflammation, together with the surgical trauma, may have triggered an osteogenetic mechanism leading to the formation of the benign tumour. It occurred inside the sinus and became clinically manifest with a classic pattern of maxillary sinus disease on the left, characterized by pain and unilateral muco-purulent rhinorrhea.

In the clinical setting, osteomas develop mainly during skeletal growth and are characterized by an independent development and slow progression related to peripheral subperiosteal bone apposition which creates a spreading, puffy shell⁵. They can develop on the edge of the bone (cortical osteoma) or in deep structures (central or medullar osteoma) and are histologically described as either eburneous, if they are composed of solid bone, or spongy, when they have gaps in the marrow of irregular structural arrangement. Both types can be situated in the maxillary sinus, but the eburneous form tends to be frontal while the spongy version is usually ethmoidal. A distinction should be made between these osteomas and displastic bone lesions, such as hyperostosis, esostosis and osteophytosis, but the non-exclusive pathology makes differential diagnosis difficult^{5,11}.

As it spreads gradually outwards, the osteoma can produce marked deformities of the face, neuralgia and nasal and paranasal inflammatory processes, but if it develops mainly inside the sinus it may not be perceived for a long time. Diagnosis is usually made by chance, usually during X-ray examination of the skull (in 40% of cases) due to facial pain or headache, especially if the site of the osteoma is fronto-ethmoidal or sphenoidal¹¹. In fact, this symptom occurs in over 60% of osteomas of the frontal sinus^{2,11}.

Radiologically, the osteoma is seen with a more or less homogeneous radio-opaque image, attached to the sinus wall with a small stem. Spiral CT scan of

the mid-face bony structures can provide useful information for planning the treatment strategy.

Whether surgery is to be the therapy of choice depends on the size of the osteoma, any worsening of symptoms and the possible onset of infection and/or complications due to compression. Osteomas are usually removed either *in toto* or broken up and sucked out under direct control.

Removal can be difficult and incomplete, with possible recurrence of the tumour, when performed using endoscopic methods, especially in the case of solid maxillary sinus tumours. Recurrence is characterized by reappearance of the symptoms, particularly pain in the face and repeated episodes of sinusitis. Early peri- and post-operative complications are mostly related to fronto-ethmoidal intervention and consist in the expulsion of cephalorachian liquid and hematomas.

Furthermore, relapse at a distance in time, ever after several years, is not uncommon. Recovery may need to be confirmed at endoscopy and imaging techniques.

In conclusion, in our opinion, osteomas of the craniofacial area are, in most cases, not recognized and are often mistaken for pathological processes of a different nature. In particular, osteomas of the maxillary sinus may be recognized after a standard head X-ray or emerge as a result of sinusitis-like symptoms. In the latter case, surgical treatment is necessary to avoid the risk of short- or long-term complications.

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