Abstract. – OBJECTIVE: Osteomas are benign tumors that frequently affect the cranio-facial region, especially the temporal bones, jaw and sinus. This lesion very rarely involves the maxillary bones. The aim of our study is to describe our surgical case series and to evaluate the diagnosis and management of peripheral craniofacial osteomas with a review of the literature.

PATIENTS AND METHODS: We retrospectively analyzed a series of 14 patients that underwent surgery for the removal of a cranio-facial osteoma, 10 cases were peripheral osteoma of the lower jaw and 4 were peripheral osteomas of the upper jaw. The 14 patients included 8 females and 6 males, with a mean age of 42 years. The median follow up period was 48 months.

RESULTS: All patients received a total surgical removal and we did not have any intraoperative complications with optimal cosmetic and functional results. Pain resolved in all cases and a single case postoperative dysesthesia occurred. No recurrence has been detected at last follow-up visit.

CONCLUSIONS: Osteomas must be well identified and differentiated from other solid diseases of the bone and should be treated if symptomatic. The elective treatment is surgical removal, resulting in a complete resolution of the pathology.

Key Words: Craniofacial osteoma, Peripheral osteoma, Gardner’s syndrome.

Facial osteomas: fourteen cases and a review of literature

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Introduction

Osteomas are benign tumors that could affect the cranio-facial region, especially the temporal bones, jaw and sinuses. This tumor may consist of a composed, compact or cancellous bone cells, being solitary or multiple, especially when presenting into Gardner’s syndrome1. Usually 3 types of osteomas could be identified: (1) the central osteoma arising from the endosteum; (2) the peripheral osteoma deriving from the periosteum and (3) the extraskeletal soft tissue osteoma, which usually develops within the muscles2. Clinically most of the lesions are asymptomatic, but in some cases they can cause pain, trismus (when also the nerve is involved) limited mandibular movements, occlusion disturbances, and facial asymmetry, especially when the mandibular condyle is involved3. Usually, an osteoma is diagnosed when it appears clearly visible at inspection or it can be incidentally discovered at Opix or facial CT exams. At radiological exams this lesion figures out as an oval radiopaque mass, well-circumscribed, attached by a broad base or pedicle to the bone cortex (mushroom-like mass)5,6. Hystological classification differentiates between two types of osteomas: compact osteomas or “ivory” are made of mature lamellar bone and they do not harvest any fibrous component. On the other side, trabecular osteomas are composed of cancellous trabecular bone with bone-marrow surrounded by a cortical bone margin5,10. In the pertinent literature there have been described 132 cases of osteomas of the craniofacial region and most of these were localized in the mandibular or frontal regions, whereas appeared to be a rare entity in maxillary bones5-10.

The aim of our study is to describe the variable clinical presentation, diagnosis and management of a series of 14 cases that underwent surgical removal of an osteoma of the maxillary bones, along with a review of the literature.
Patients and Methods

We analyzed a series of 14 patients, 8 women and 6 men with a mean age of 42 years (range 26 and 64 years), with an osteoma of the cranio-facial region, treated surgically at the Maxillofacial Surgery Department of the University Federico II of Naples, between May 2000 and March 2010. The series consisted of 10 peripheral osteomas of the lower mandibular bone and 4 were peripheral osteomas of the maxillary bone (Table I). All patients complained of local pain. Opx and a CT were performed to rule out the diagnosis. Pathological report confirmed diagnosis of osteomas lining out 9 cases of compact hysto-type and 5 of cancellous hystotype. Mean follow-up was of 48 months (range). We herein report the three most representative cases.

Results

All patients underwent intraoral approach for the removal of an osteoma of the maxillary bones; total removal of the lesion was achieved in all cases. Of the 14 cases of our series, 3 involved the mandibular angle area, 7 the anterior body, 4 the alveolar processes. Pain completely resolved immediately after surgery in all cases, as well as all mouth opening functional limitations. Nine patients out of the eleven (9/11, 81.8%) presenting preoperatively facial swelling had complete cosmetic and functional recovery, whilst all patients with facial asymmetry had complete restoration. We did not report any intraoperative complication, whereas transient dysesthesia of the right V3 branch of the trigeminal nerve occurred postoperatively in a single case (1/14, 7.14%) presenting osteoma of the right mandible angle area. There were no recurrences at last follow up visit of 4 years.

Illustrative Cases

Case 1

A 26-year-old male patient was complaining of pain in the right half mandible since 5 years and presented swelling of the right posterior cheek. An Opx and a CT scan (Figure 1 A-B) showed the presence of a thickened bony area occupying the right mandibular angle. There were no symptoms or neurologic defects related to the lesion and there were no other lesions in the facial or cranial bones. The patient underwent surgical removal of the neoplasia. The surgical approach accounted on an intra-oral route in order to maximize the cosmetic result (Figure 1 C). An immediate postoperative Opx (Figure 1 D) confirmed the complete removal of the pathology. Two days after surgery dysesthesia along the right V3 branch of the trigeminal nerve occurred: steroid therapy along with B group vitamins support was administered and after two months symptoms totally relieved. Pathology report disclosed a diagnosis consistent with an osteoma of compact type.

Case 2

A 54 yo male patient noted the presence of two lesions growing in right maxilla and started complaining of pain seven months prior to hospit-
tal admission; no facial asymmetry was clinically visible. Suddenly, he noted increase of thickness on both sides of the upper jaws. CT scan showed two hyperdense lesions at level of lateral arches of the upper jaws (Figure 1 A-B). Both lesions (Figure 2 C-D) were completely removed via an intraoral approach at the same time (Figure 2 E-F). Histological examination showed osteoma of compact type for both. The postoperative course was uneventful and at the 48-month follow up the areas healed.

Case 3
A female patient, aged 26, was referred to our department complaining of diffuse pain at right Temporo-mandibular joint area, associated with local swelling. Opx and CT scans ruled out a bony lesion involving the outer aspect of the right ascending branch of the mandible (Figure 3 A-B). Intraoral approach was adopted to completely remove the lesion (Figure 3 C). No intra-operative complication occurred; although patient was relieved from pain immediately, facial swelling did not improve (Figure 3 D). At last follow up recurrence was not detected and swelling was noted.

Discussion
Osteoma is a benign, slow growing lesion that could affect the craniofacial region very rarely; indeed, to our knowledge, 132 cases of osteoma of the jaws have been reported in the pertinent literature3,5-36,38,40,41. Females present a higher incidence rate, with no predilection for any specific age range. Concerning the etiopathogenesis there are several theories, which received some credit along the years. It has been pointed out that they could be congenital29-31, develop as neoplastic mass or, more likely, as inflammatory lesions3,32. However, a common path underlying the developmental process of osteomas has been recognized: it has been supposed that a combination of a trauma and muscle activity can initiate an osteogenic
Figure 2. A, B, CT scan showing two hyperdense bony lesions in the upper jaw. C, D, Pre-operative image of intraoral mucosa distorted by lesions in both sides. E, F, Intra-operative image showing the exposure of the lesions; both appear exofitic tumors.
reaction. Usually these lesions are diagnosed when they become visible or figure out with local pain; on the contrary, they can be incidentally discovered at radiological examination\textsuperscript{33-35}. Differential diagnosis should take into account several inflammatory and neoplastic pathologic entities, such as exostosis, chronic focal sclerosing osteomyelitis, ossifying fibroma, chondroma, osteosarcoma, Paget’s disease, fibrous dysplasia and odontoma\textsuperscript{36}. Osteomas can be classified as central or peripheral; they occur mostly in the head and neck region, often involving the paranasal sinuses, above all the frontal sinuses. The peripheral osteomas have been described in the jaw bones, but this localization seems very uncommon. Aside from those lesions reported as entities of Gardner’s syndrome, peripheral osteomas of the jaws account on 69 cases\textsuperscript{3}. 63 of them were located in the mandibular bones. Concerning site of origin, there is a certain predilection for the mandibular body (4 cases anterior region, posterior region 19 cases), followed by the condyle (18 cases), the angle (9 cases), ascending ramus (7 cases), coronoid process (5 cases) and sigmoid notch (1 case)\textsuperscript{3}. On the other hand, of the 6 osteomas of the upper jaw, 4 were involving in the alveolar process and 2 in the hard palate\textsuperscript{3}. The patients in our series presented sim-
ilar distribution with 10 cases involving the lower jaw and only 4 the upper jaw. The most common symptom was local pain, although we noticed further symptoms such as headache, facial asymmetry, limited mouth opening and trismus, which seldom appeared in the literature. These symptoms have to be directly related to the “mass effect” of the lesion impinging vital structures. Accordingly, a flow chart for osteoma treatment could be drawn: when the osteoma determines cosmetic disfigurement, limitations or loss of functions, it shows significant volume increase, cogent symptomatology, and/or severe pain refractive to medical therapy should be treated surgically. On the other side, when asymptomatic, there is no univocal consensus in regards, with little more evidence in favor of watchful waiting. Indeed, there are no reports of malignant transformation of a peripheral osteoma. Surgical treatment should account on radical surgical removal, extended also to the surrounding normal bone, with the intraoral approach being preferred for aesthetic reasons and the extraoral reserved for those larger tumors, when a larger exposure is required. As a matter of facts, the occurrence of several complications could overburden this surgery. Indeed, in our series a very prolonged V3 branch dysesthesia occurred although the patient underwent a very accurate preoperative work-up and a cautious intraoperative dissection was carried out.

In these regards, thanks to recent technological advances, it should be advisable to have high quality CT scan prior to the surgical procedure in order to rule out pearly all anatomical spatial relationships and eventual keypoints. Furthermore, recent literature reports the advantages of the use of piezo-surgery in the dissection of these lesions. Piezo-surgery allows removal of the bone without any damaging to soft tissues and nerves, reducing post-operative pain, dysesthesia and swelling.

Finally, it should be reminded that recurrence are very rare, with a single case occurred 9 years after the surgical treatment, described by Bosshardt et al. Over a mean follow up of 48 months we did not report any recurrence.

### Conclusions

Craniofacial osteomas are more frequent in the mandible, with no predilection for any specific age range. In this area, they mainly occur in the body, condyle, angle, ascending ramus, coronoid process and sigmoid notch. These tumors must be differentiated from other diseases of the bone and should be treated surgically, when symptomatic, minding the functional and aesthetic results.

### Conflict of Interest

The Authors declare that there are no conflicts of interest.

### References


