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Paleopathological evidence of paranasal lesions: Two cases of frontal sinus osteomata from Imperial Rome

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ABSTRACT

The archaeological excavations carried out in 1999 in the Collatina necropolis of the Roman Imperial Age (1st–3rd centuries AD) (Rome, Italy) discovered the skeletal remains of two adult males with evidence of paranasal lesions. Both individuals showed *postmortem* damage in the frontal bone, through which it was possible to macroscopically detect an oblong new bone formation. In both specimens, radiological examination of the defects' morphology showed new pediculated-based bone formations. Radiology also confirmed the presence of benign osseous masses arising from the right frontal sinus and interpreted as osteomata. Their dimensions did not exceed 10 mm, so that mechanical complications and compression of the adjacent structures could be ruled out. The osteomata of paranasal sinuses are rarely reported in paleopathology, since they can be discovered only incidental to bone breakage or radiography. Hence, the evaluation of their occurrence in past populations represents an important challenge. The two cases presented here show direct and rare evidence of frontal sinus osteomata dating back to the Roman Imperial Age.

1. Introduction

The osteoma is the most common slow-growing benign osteogenic lesion largely affecting the skull and the paranasal sinuses. Morphologically, it appears as a sessile mass attached to the bone surface or as a pedunculated-based mass; it may be single or occasionally multiple, as in Gardner's syndrome (Alexander et al., 2007). Clinical data suggest that the frontal sinus is the most affected of the paranasal sinuses (71.8% of cases), followed in descending order by the ethmoid (16.9%), maxillary (6.3%) and sphenoid (4.9%) sinuses (Sanchez Burgos et al., 2013). An osteoma may grow on the cranial vault in the form of a bone exostosis and more rarely from the inner cranial table (Aufderheide and Rodríguez-Martín, 1998). When the lesion grows on the bone tissue, it is called homoplastic; if it occurs on other tissues, it is called heteroplastic (Hakim et al., 2015).

The current medical literature (Rokade and Sama, 2012) suggests that peak incidence of paranasal sinus osteomata fall between the 3rd and 4th decades of life, with a male predominance (M:F ratio 1.5:1 to 3.1:1).

Histologically, three types of osteomas can be differentiated on the basis of the proportions between compact and cancellous bone (Rokade and Sama, 2012): 1) the ivory or eburneated osteoma, typical of the skull vault (the so-called "button osteoma"), composed of dense mature lamellar bone without Haversian systems; 2) the mature osteoma or osteoma *spongiosum* composed of trabecular, mature lamellar bone with Haversian system; and 3) the mixed osteoma, a mixture of the ivory and mature types.

As suggested by McHugh et al. (2009), an osteoma of the paranasal sinuses rarely exceeds 20 mm, thus resulting mostly asymptomatic and fortuitously diagnosed by x-ray in 0.43% of patients, and by computed tomography (CT) in 3% (Rokade and Sama, 2012). Nevertheless, it may increase in size resulting in giant paranasal sinus osteoma (Sanchez Burgos et al., 2013; Exley et al., 2015), leading to several complaints such as headaches, acute or chronic sinusitis, mucocele formation, brain abscess, diplopia, proptosis, epiphoria, intracerebral intrusion, protrusion or intrusion in the orbital spaces, and facial or cranial deformities with cosmetic implications (Summers et al., 2001; Sinha et al., 2003).

The etiology of osteomata is strongly debated today. The three

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dominant theories include (Dell'Aversana Orabona et al., 2015) the embryologic theory, the traumatic theory, and the infectious theory. The embryologic theory suggests that osteomata are the result of developmental anomalies arising in the fronto-ethmoid region, where membranous and cartilaginous tissues are in contact during embryonic life. However, the embryologic theory does not explain the large number of osteomata located far from the fronto-ethmoid junction. The traumatic theory identifies a previous trauma as a possible etiologic event, but it does not explain the large number of patients with no prior history of trauma. The infectious theory originally hypothesized that osteomata are precipitated by sinus infections.

In paleopathology, the identification of paranasal sinus osteomata is rather uncommon, because osteomas are casually detected when skeletal remains are submitted to radiological examination for other research purposes or when the skull is broken and the osteoma is directly visible, as in the cases of our study. For these reasons, a review of the paleopathological literature counts a very limited number of cases of paranasal sinus osteomata (Kindler, 1972; Bourry et al., 1990; Yniguez, 1994; Hagedorn et al., 2002; Rühli et al., 2004; Premužic et al., 2013). Therefore, these two Italian cases dating back to the Roman Imperial Age can expand our knowledge of such lesions in the past.

2. Materials and methods

The skeletal remains were discovered in the Collatina necropolis (eastern part of modern Rome) in 1999, during the archaeological excavations directed by the Archaeological Superintendence of Rome (Buccellato et al., 2008) (Fig. 1). Over 2500 burials were found and were mainly from the urban population living in Rome during the Imperial Age (1st-3rd centuries AD).

The two skeletons (code numbers SerM_200 and Bas_460) were supine in simple graves excavated in the tuff rock, with one iron nail in each tomb as apotropaic grave good. The tomb of Bas_460, covered with tiles, also contained a small clay pot. Both skeletons were in anatomical position. SerM_200 was complete and well-preserved with class 5 Anatomical Preserved Index (API) and class 5 Qualitative Bone Index (QBI); Bas_460 was incomplete and in poor condition, with class 4 for API and class 3 for QBI (Bello et al., 2006).

The sex estimation was based on the morphological characteristics of the skull and pelvis (Buikstra and Ubelaker, 1994). Age determination was based on dental wear (Lovejoy, 1985), on the stage of fusion of the cranial sutures (Meindl and Lovejoy, 1985), on the changes of the auricular surface (Lovejoy et al., 1985), and on the pubic symphysis of the pelvis (Brooks and Suchey, 1990). Pathological evidence was evaluated by using a lens with low magnification (5X) and consulting the main paleopathological literature (Ortner, 2003; Aufderheide and Rodríguez-Martín, 1998). Macroscopic observation was followed by imaging study. Direct Cone Beam Computed Tomography (CBCT) equipment (PlanMeca Promax Classic 3D) was used to obtain 3D acquisition (parameters 5.6–12 mAS with 86–85 kV), and 2D acquisition (parameters 10–10.5 mAS with 64 kV).

3. Results

3.1. Bas_460

The skeleton was almost complete, but the skull was poorly preserved. The morphological features of the pelvis and skull indicated that the individual was a male, 40–50 years of age at death on the basis of dental wear and ectocranial suture closure. The *postmortem* fractured frontal bone allowed to observe a new small ovoliform bone formation in the right frontal sinus (Fig. 2). Radiological examination confirmed the presence of a new well-circumscribed lobulated bone mass protruding from the central portion of the right frontal sinus and measuring 8.2 mm in length and 4.4 mm in width. The mass exhibited a ground-glass appearance related to the trabecular structure of the bone tissue (Fig. 3). No other lesion in the skull was detected, including those referable to maxillary sinusitis.

3.2. SerM_200

The skeleton, well preserved and complete, belonged to an adult male as indicated by the morphology of the skull and pelvis, aged 40–50 at the time of death on the basis of the changes of pubic symphysis and auricular surface of the pelvis, and on dental wear pattern. The right portion of the frontal bone, in correspondence to the frontal sinus, showed triangular *postmortem* damage, which exposed a small pear-shaped bone protuberance with smooth surface (Fig. 4). Radiological examination confirmed the presence of a homogeneous fully radiodense new bone mass, composed exclusively of compact bone. The pedunculated lesion, 4.6 mm long and 3.3 mm wide, was projected from the central portion of the right frontal sinus (Fig. 5). No other lesion in the skull, including those associated with maxillary sinusitis, was detected.

4. Differential diagnosis

The pathologic features of the paranasal sinus lesions suggested that the differential diagnosis should concentrate on the following conditions: osteoma, osteoid osteoma, osteoblastoma, and ossifying fibroma. Osteoid osteomata and osteoblastomas are rare benign bone neoplasias that predominantly affect the vertebral column and the long bones of young males, while the skull and paranasal sinuses are seldomly involved (Caltabiano et al., 2012; Pelargos et al., 2015). Both osteoid osteoma and osteoblastoma consist of a nidus of osteoid and woven-bone rimmed by a perifocal bone sclerosis. Their differentiation is merely based on a dimensional criterion for which osteoma osteoid rarely exceeds 1.5 cm, while an osteoblastoma typically exhibits a larger nidus (> 2 cm) (Unni and Inwards, 2010). Ossifying fibroma are fibro-osseous lesions that typically occur during young adulthood, affecting the long bones, the mandible and paranasal sinuses with slight predisposition for females (M:F ratio 1:1.04, n = 55) (Manes et al., 2013). Radiographically, ossifying fibroma appears as a sharply circumscribed radiolucent round or oval lesion composed of woven bone surrounded by an eggshell of bone sclerosis (Manes et al., 2013). The radiological appearance and the tissue features of osteoid osteoma, osteoblastoma, and ossifying fibroma were not observed in our two specimens.

Malignant lesions can also be ruled out since our cases did not display the typical characteristics of malignant neoplasms such as inhomogeneous appearance, hazy margins, osteolytic changes and cortical expansion. The two lesions observed in the Roman samples consisted of well-defined new bone formations composed of mature and dense lamellar bone with smooth surfaces and with no marrow spaces. These features, associated with cranial localization, led to a final diagnosis of osteomata of the frontal sinus. Specimen SerM_200 was entirely composed of dense mature compact bone, lacking bone trabeculae and similar to an ivory osteoma, while specimen Bas_460 was classified as a mature or *spongiosum* osteoma on the basis of its radiological ground glass appearance correlated to the trabecular structure of the bone tissue (Dell'Aversana Orabona et al., 2015). To the best of our knowledge, our Italian case represents the first case of *spongiosum* osteoma ever documented in archaeological records, since all the cases published so far concern osteomas of the compact type (Table 1).

5. Discussion and conclusions

Osteomas are considered the most common slow-growing benign lesions, but, as suggested by Sadry et al. (1988), spongious osteomas grow more rapidly than the compact ones. Clinical data concerning the general growth rate estimate 0.79 mm/years in cephalocaudal (CC) and 0.99 mm/years in mediolateral (ML) dimensions (Buyuklu et al., 2011).