Sino-Orbital Osteoma
A Clinicopathologic Study of 45 Surgically Treated Cases With Emphasis on Tumors With Osteoblastoma-like Features

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Context.—Osteomas are limited almost exclusively to craniofacial and jaw bones. Histologically, they can be divided into ivory, mature, or mixed types. Osteomas may have osteoblastoma-like areas and distinguishing it from true osteoblastoma can be challenging. Some believe osteomas with osteoblastoma-like features behave more aggressively.

Objective.—To perform a clinicopathologic comparison of sino-orbital osteomas both with and without osteoblastoma-like features.

Design.—We studied 45 surgically excised sino-orbital osteomas. Tumors were categorized as ivory, mature, or mixed type and presence of osteoblastoma-like areas and Paget-like bone were noted. Clinical features of those with and without osteoblastoma-like areas were compared.

Results.—Men outnumbered women (3:2); median age was 37 years. Frontal sinus was the most common location (62%) followed by ethmoid and maxillary sinuses. Twelve tumors (27%) involved the orbit, 2 primarily and 10 secondarily. All cases were symptomatic with headache, sinusitis, visual changes, pain, and proptosis being most common. Seventeen tumors (38%) had osteoblastoma-like areas. Extension into an adjacent sinus/anatomic compartment was more common in osteoblastoma-like tumors (47% versus 29%), including more frequent orbital involvement (41% versus 13%). Visual changes were more frequent in the osteoblastoma-like group. Distribution of histologic subtypes and Paget-like bone were similar between the 2 groups. Osteomas with osteoblastoma-like features were more often incompletely excised (25% versus 14%). However, clinical recurrence was less common (8% versus 27%).

Conclusions.—Osteoblastoma-like features are common in sino-orbital osteomas, but it does not correlate with more adverse clinical features or worse outcome. Osteoblastoma-like areas appear to represent active remodeling within an osteoma rather than defining a distinct clinicopathologic entity. Distinguishing it from osteoblastoma may require careful histologic evaluation and radiographic correlation.

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Osteomas are benign, generally slow-growing, bone-forming tumors limited almost exclusively to craniofacial and jaw bones. They can be subdivided into bone surface tumors (or exostoses) that primarily involve the cranial vault, mandible, and external auditory canal and the more common sino-orbital (or paranasal sinus) osteomas that arise from bones that define the paranasal sinuses, nasal cavity, and orbit. Frontal, ethmoid, maxillary, and sphenoid sinuses are most frequently affected in that order.1 Sino-orbital osteomas present with clinical findings due to mass effect such as pain, headache, facial distortion, proptosis, and visual changes or due to nasofrontal duct obstruction by frontal sinus tumors, which cause mucopurulent discharge and infections. Tumors that erode the supraorbital plate and dura mater may cause cerebrospinal fluid leakage, pneumatocele, meningitis, or cerebral abscess.

Histologically, although the surface exostoses are usually formed of compact bone, sino-orbital osteomas are composed of variable amounts of compact and cancellous bone. Fu and Perzin2 subdivide them into ivory and mature types depending on the proportions of dense and cancellous bone. However, this separation is not thought to be clinically relevant and several overlapping classification schemes are reported in the literature and bone pathology textbooks with no universally accepted nomenclature (Table). ivory osteomas are composed of dense, mature, lamellar bone with little fibrous stroma. Mature osteomas are composed of large trabeculae of mature, lamellar bone with more abundant fibrous stroma. Mature osteomas have osteoblastoma-like features by Dorfman and Czerniak.3 In addition to potentially being mistakenly diagnosed as osteoblastoma or osteoid osteoma, it has been suggested they be...
have more aggressively than traditional sino-orbital osteomas with locally destructive growth and recurrences. However, very little data exist in the literature to substantiate this claim.

The clinical and radiographic features of sino-orbital osteomas have been well documented in numerous studies. However, very few studies detail the pathology. Thus, the purpose of this study was to review our experience with a series of surgically excised osteomas, documenting the clinicopathologic and radiographic findings and paying particular attention to osteomas with osteoblastoma-like features.

**DESIGN**

This study was approved by the institutional review board at the University of Michigan Health System. Forty-five consecutive cases of sino-orbital osteoma were identified from the files of the Department of Pathology between March 1986 and January 2008. Patients with surface osteomas (exostoses) of the frontal bone (10 patients) and external auditory canal (3 patients) were excluded from this study. In addition, during the study period, 4 patients were diagnosed with craniofacial osteoblastomas. On review of the clinical, radiologic, and pathologic material, 2 of these were felt to be osteomas with osteoblastoma-like features. The reason for the change in diagnosis in both cases was the presence of a polypoid mass arising from the bones of the orbit/sinus that were considered osteomas on initial review of the imaging studies. Histologically, both were composed predominantly of dense mature bone associated with small foci with osteoblastoma-like features. Regarding the 2 osteoblastomas, 1 involved the maxillary alveolus with involvement of the adjacent palate bone and 1 involved the mandibular body in the region of the inferior alveolar nerve. On imaging, both involved the medullary compartment and had imaging findings that were consistent with osteoblastoma. The patient with the mandibular lesion underwent excision of recurrent/persistent disease 7 months after diagnosis but both patients were eventually lost to follow-up. Follow-up data for the remaining 45 patients were obtained from medical records and the referring clinicians where appropriate.

All histologic sections from each case were reviewed to confirm the diagnosis. Tumors were subclassified as ivory type, mature type, or mixed ivory/mature type according to the predominant histologic components. Ivory type was defined as an osteoma composed predominantly of dense mature lamellar bone with little medullary stroma. Mature type was defined by large trabeculae of mature lamellar bone with or without osteoblastic rimming and separated by paucicellular fibrous stoma. Mixed ivory/mature type was defined as a tumor with at least 1 low-power field (×40 magnification) each of ivory and mature patterns.

In addition, the presence of osteoblastoma-like areas and Paget-like bone were tabulated. Osteoblastoma-like areas were defined as discrete areas composed of haphazardly arranged, interanastomosing woven bone trabeculae lined by plump osteoblasts and associated with a cellular loose fibrous stroma containing numerous small vessels. The largest linear dimension of the osteoblastoma-like area was measured from the slides and tabulated. Paget-like bone was defined as bone with irregular cement lines resulting in a characteristic mosaic pattern.

These findings were then correlated with clinical presentation, radiographic description, operative report, and follow-up information. Specifically, we recorded the anatomic localization, presenting signs and symptoms, presence of extracavitary extension, recurrence(s), and patient status at last available follow-up. For the purposes of this study, extracavitary extension was defined as extension from the tumor epicenter into an adjacent structure (usually an adjacent paranasal sinus or orbit) as determined radiologically or surgically. We then compared tumors with and without osteoblastoma-like features for clinical and prognostic differences.

Available imaging studies were reviewed by a radiologist with expertise in neuroradiology and head and neck radiology (S.K.M.). Ten patients had available imaging studies performed prior to definitive surgery, 4 with and 6 without osteoblastoma-like features. The imaging studies were evaluated to determine if the computed tomography (CT) findings correlated with the histologic findings.

**RESULTS**

**Clinical Presentation**

Forty-five patients underwent surgical excisions for symptomatic sino-orbital osteomas during the study period yielding an average of 2 cases per year. The study population consisted of 27 men and 18 women (male to female ratio, 3:2) with median age of 37 years (range, 12–70 years). Three patients were known to have Gardner syndrome.

Frontal sinus was the most common primary site (62%), followed by ethmoid sinus (24%). Three tumors (7%) involved frontal and ethmoid sinuses concomitantly making it impossible to determine a single site of origin. Maxillary sinus was the primary site in 4 cases (9%) and the orbit was the primary site in 2 cases (4%). Seventeen tumors (38%) had extracavitary extension at the time of diagnosis, including 10 (22%) with secondary orbital involvement.

All patients were symptomatic with the most common signs and symptoms being headache (51%), visual changes (36%), sinusitis (29%), pain (24%), and proptosis/globe displacement (24%). All but 1 patient, who presented with mass alone, had at least one of these 5 signs and symptoms. Other clinical findings were mucocele, abscess, nasal polyphs, rhinorrhea, epistaxis, periorbital edema, ptosis, cerebrospinal fluid leakage, meningitis, and dizziness. Most patients had multiple symptoms. In patients in whom du-
ration of symptoms was reported (n = 21), the median time was 12 months.

**Pathology**

Size determinants of the in-situ tumor based on CT findings or of the resected tumor specimen were available in all but one of the patients. The gross tissue measurement was considered less reliable due to the fragmented nature of many of the resected specimens. The mean size of the tumors, whether intact or fragmented, was 3.1 cm with a median of 3.0 cm (range, 0.6–8 cm).

Histologically, the mixed ivory/mature type was seen most often (60%) followed by the mature (29%; Figure 1) and ivory types (11%; Figure 2). Seventeen specimens (38%) had areas with histologic features indistinguishable from osteoblastoma (Figure 3). Osteoblastoma-like areas were typically present at the base of the tumor (ie, at the site of attachment to the bone of origin) and ranged in size from 0.5 to 1.9 cm (median size, 1.0 cm). Dense mature bone defined the periphery where it had a rounded, sharply demarcated surface lined by compressed respiratory epithelium. Microscopically, the osteoblastoma-like areas were composed of narrow interanastomosing trabeculae of woven bone rimmed by osteoblasts and osteoclasts (Figure 4). The interosseous stroma was loosely textured and edematous, and composed of fibroblasts and numerous thin-walled, often ectatic and congested vessels. No tumors had epithelioid osteoblasts characteristic of the aggressive (epithelioid) variant of osteoblastoma. In 4 tumors, osteoblastoma-like areas appeared multifocal, consisting of more than one focus separated by thick mature bone. With intact specimens, osteoblastoma-like areas were grossly visible as brown, gritty, finely trabeculated zones (Figure 5). Paget-like bone was present in 42% of tumors, evenly distributed between the osteoblastoma-like and non–osteoblastoma-like tumors (Figure 6).

**Clinicopathologic Correlation**

Men outnumbered women in the osteoblastoma-like group (3:1) as compared with those without osteoblastoma-like features (1:1). The median age was similar between the 2 groups (37 versus 39 years). Of the 3 patients with Gardner syndrome, 2 had tumors with osteoblastoma-like features.

Similar to the study population overall, tumors with osteoblastoma-like features occurred most frequently in the frontal (59%) and ethmoid sinuses (44%). No tumor with osteoblastoma-like features occurred most frequently in the maxillary sinus (59%) and ethmoid sinuses (44%). Similar numbers of tumors in the 2 groups had Paget-like bone; 47% in the group with osteoblastoma-like features and 39% in the group without.

Follow-up information was available for 35 patients (78%) including 13 of 17 (76%) of those with osteoblastoma-like features and 22 of 28 (79%) of those without. Mean overall follow-up time was 61 months (range, 2–468 months). Mean follow-up differed between the 2 groups (47 versus 69 months); however, when 1 patient in the non–osteoblastoma-like group with 39 years of follow-up was not included, follow-up times were very similar (47 and 50 months, respectively). Patients with osteomas with osteoblastoma-like features had a lower clinical recurrence rate than those without (8% versus 27%). In addition, 3 patients (25%) with osteoblastoma-like features and 3 patients (14%) without were initially incompletely excised with gross residual tumor remaining after surgery. Patients with osteomas with osteoblastoma-like features were more likely to be alive with disease than those without (31% versus 18%). No patient has died from the osteoma.

**Radiologic Review**

The imaging findings of the ivory type of osteoma were characterized by lesions composed predominantly of very dense bone with only small lucent areas at most (Figure 7). The mature type tended to have areas of very dense bone of varying thickness, adjacent to less dense areas with the appearance similar to the fibrous matrix often seen in fibrous dysplasia or other fibro-osseous lesions (Figure 8). The appearance of the osteoblastoma-like group was often characterized by a central area of a less dense fibrous matrix at the base surrounded by a periphery of very dense bone that was thinner compared with the other types (Figure 9). These CT findings appear to correlate with the histologic findings; however, there was radiologic overlap, especially among the mature, mixed, and osteoblastoma-like groups.

**COMMENT**

Osteoma is the most common tumor of the paranasal sinuses. The exact incidence, however, is unknown because small, clinically silent osteomas are commonly detectable by CT scan in up to 3% of the population. Symptomatic osteomas are typically associated with headache, visual changes, facial pain, and sinusitis. It is generally agreed that symptomatic sino-orbital osteomas should be surgically removed as well as those that continue to enlarge and those that extend beyond the boundaries of the sinus. Because osteomas are benign, slow-growing tumors, surgery is aimed at reduction of symptoms by local excision as opposed to radical deforming resection. Thus, tumors are not always completely removed. The management of asymptomatic sino-orbital osteomas is not well established.

The most common locations are frontal, ethmoid, maxillary, and sphenoid sinuses in that order. Osteomas may...
Figure 1. Typical mature-type sino-orbital osteomas were composed of broad trabeculae of mature lamellar bone without significant osteoblastic rimming. The stroma was characteristically paucicellular with spindled to stellate fibroblasts/myofibroblasts embedded within a collagenous stroma (hematoxylin-eosin, original magnification ×100).

Figure 2. Most osteomas grew in an expansive fashion extending close to and compressing the adjacent paranasal sinus mucosa (arrowhead). Ivory-type sino-orbital osteomas were composed predominantly of dense mature lamellar bone with little fibrous medullary stroma (hematoxylin-eosin, original magnification ×100).

Figure 3. Low-power image from a typical osteoma with osteoblastoma-like features. Most had an outer rim of dense (ivory-type) lamellar bone that gradually transitioned to the osteoblastoma-like focus (asterisk). This transitional area varied from abundant (×40 field) to focal and the size and number of the osteoblastoma-like areas varied as well (hematoxylin-eosin, original magnification ×40).
also primarily involve the orbit and nasal cavity. Radiographically, osteomas typically manifest as homogeneously calcified, lobulated, sharply defined tumors that tend to form polyloid intracavitary growths. Most are less than 2 cm but can sometimes become massive, referred to as giant osteomas.13,15 Osteomas may have central areas of radiolucency that in some cases correlate with woven bone with microscopic features indistinguishable from osteoblastoma.3,10

The clinical findings in our series of surgically treated osteomas parallel those in the literature. Men outnumbered women by 50%, median age was 37 years, frontal sinus was the most common location, and all patients were symptomatic with headache, visual changes, sinusitis, pain, and proptosis/globe displacement being the most frequent symptoms. Six patients had recurrent disease that required reexcision, some of whom had multiple operations. Six patients had persistent asymptomatic tumor following partial excisions.

Based on the criteria of Fu and Perzin,2 11% of our tumors were ivory-type, 29% were mature-type, and 60% were mixed ivory/mature-type osteomas. Paget-like bone, an indicator of active remodeling, was a common finding, being focally identified in 42% of our tumors. Areas indistinguishable from osteoblastoma were identified in 17 tumors, accounting for a significant percentage of our cases (38%). The amount of osteoblastoma-like tissue was variable among cases ranging from small isolated foci to quite extensive and appeared multifocal in 4 tumors. There was a distinct zonation in these tumors with the osteoblastoma-like areas localized to the base of the osteoma while thick mature bone formed the periphery.

The clinical behavior of our cases of osteoma with osteoblastoma-like features paralleled those of conventional osteomas in symptomatology and incidence of recurrent and persistent disease. Although patients with tumors with osteoblastoma-like features were more likely to have extracavitary extension (47% versus 29%) and to be alive with persistent/residual tumor compared with those with conventional tumors (33% versus 18%), they were less likely to have clinically recurrent disease (8% versus 27%). Thus, these tumors do not appear to represent a more aggressive form as has been suggested.3

Distinguishing osteoma with osteoblastoma-like features from other bone-forming and fibro-osseous lesions in the craniofacial and jaw bones can be challenging and frequently requires radiologic correlation. However, based on the histology alone, osteoid osteoma and osteoblastoma would be the major differential diagnostic considerations. In contrast, osseous fibroma and fibrous dysplasia would less likely be confused with this entity as the latter lesions have a more prominent collagenous spindle cell stroma. Osteoid osteomas are very rare in the craniofacial and jaw bones, accounting for only 1% of tumors.9 We are aware of only 2 reported cases involving the paranasal sinuses.16,37 It is distinguished from osteoblastoma only by size with a nidus usually less that 1 cm,18 because the histologic features are essentially indistinguishable. Unlike osteomas, osteoid osteomas have a distinctive pain pattern and should not form an ossified intracavitary polyloid mass.

Osteoblastoma is most commonly located in the vertebral and long bones. However, it occurs in virtually any bone including the craniofacial and jaw bones.18 Mandible is a relatively common location for osteoblastoma18 where it is often associated with the root of a tooth and referred to as cementoblastoma.19 Less frequently it involves the cranium or maxilla, which account for only 4% and 3% of cases, respectively.18 Primary osteoblastoma of the paranasal sinuses, however, is rare. We are aware of only 23 published cases, all single case reports,2,20–41 many of which we believe actually represent osteomas with osteoblastoma-like features.

Microscopically, although focally indistinguishable from osteoblastoma, osteoma with osteoblastoma-like features has much more mature bone in the form of solid/compact (ivory osteoma) and dense cancellous (mature osteoma) bone. In addition, the outer contour of an osteoma has a smooth rounded surface, often lined by respiratory mucosa, representing the outer surface of its polyloid growth within the sinus cavity. True osteoblastoma, by contrast, will form an expansile intramedullary or periosteal bone tumor with a radiolucent or partially calcified appearance, often with surrounding sclerosis.19 In fact, several of the reported cases of osteoblastoma of the paranasal sinuses may indeed represent osteomas with prominent osteoblastoma-like features based on the radiographic illustrations.2,3,20

Although, limited, our radiologic review revealed that CT findings correlated well but not perfectly with the histologic subtype, because there were overlapping radiologic findings among the subtypes. The primary imaging finding of the ivory type was a predominance of very dense bone. However, there were often small foci of more lucent areas suggesting that the final histologic subtype may have been affected by sampling. The mature type of osteoma consisted of a more equal distribution of very dense and less dense bone suggesting a fibrous component. Finally, osteomas with osteoblastoma-like features tended to have larger areas of less dense bone with a peripheral rim of dense bone.

In summary, areas indistinguishable from osteoblastoma are common in sino-orbital osteoma, the extent of which is variable. Tumors with osteoblastoma-like fea-

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**Figure 4.** High-power image from an osteoblastoma-like area demonstrating the characteristic woven bone rimmed by abundant osteoblasts and scattered osteoclasts. The stroma was characterized more cellular than the mature-type stroma and contained numerous dilated, thin-walled vascular channels (hematoxylin-eosin, original magnification ×200).

**Figure 5.** Typical gross specimen of an osteoma with osteoblastoma-like features. The expansile bony mass can be seen compressing the overlying sinus mucosa (arrowheads). The dense lamellar bone appeared chalky white and tended to be located adjacent to the compressed mucosa while the more vascular osteoblastoma-like area (asterisk) was brown and tended to be located at the attachment to the bone of origin.

**Figure 6.** Paget-like bone demonstrating the characteristic irregular reversal lines secondary to bone remodeling and resulting in a mosaic pattern similar to that seen in Paget disease, which was a common finding in both osteoblastoma-like and nonosteoblastoma-like tumors (hematoxylin-eosin, original magnification ×200).
tures do not appear to behave more aggressively than those without osteoblastoma-like areas as has been previously suggested. Thus, it does not appear to be a distinct clinicopathologic entity. Instead, osteoblastoma-like areas most likely represent foci of bone remodeling, as was suggested by Bullough in his seminal article and as suggested by the frequent presence of Paget-like bone in the current study. Distinguishing osteoma with osteoblastoma-like features from true osteoblastoma can be challenging in the absence of radiographic correlation. However, unlike osteoblastomas, which form expansile, partially calcified tumors, osteomas with osteoblastoma-like features form heavily ossified masses with a less radio-dense center that tend to grow as intracavitary polypoid tumors.

References