Sino-Orbital Osteoma With Osteoblastoma-Like Features

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Abstract An 18 year old male presented with worsening headaches, pain with ocular movement and swelling that involved the left anterior periorbital and frontal sinus region. Radiographic images revealed a polypoid bony mass of mixed radiodensity extending into the left and right frontal sinuses. Histologic examination of the resection material resulted in the diagnosis of an osteoma with osteoblastoma-like features, an osteoma variant that has zones indistinguishable from an osteoblastoma. The clinical, radiographic, and morphologic features of sino-orbital osteoma with osteoblastoma-like features are discussed.

Keywords Osteoma · Osteoblastoma-like · Sino-orbital · Frontal sinus · Paranasal sinuses · Radiology · CT

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History

An 18 year old Hispanic male presented with worsening headaches, pain on ocular movement, and swelling that involved the left anterior periorbital and frontal sinus region. He reported feeling increased sinus pressure during the preceding 30 days that did not resolve after a 10-day regimen of antibiotics. He had a past history of “worrisome” sinusitis. The primary diagnostic concern, prior to imaging, was chronic sinusitis and secondary periorbital cellulitis with extension into the orbit.

Radiographic Features

Conventional CT images revealed an expansile, polypoid mass of mixed radiodensity extending into the left frontal and right medial frontal sinuses with additional projection posteriorly into frontoethmoidal recess (Figs. 1, 2, 3). Residual left frontal sinus airspace and left ethmoid air cells were opacified. The left frontoethmoidal recess and left anterior ethmoidal air cells showed dense, partially calcified soft tissue material, which also appeared to involve the anterior left lamina papyracea (orbital wall) (Fig. 4). The lobulated bony mass featured a partial rim of thickened compact, hyperdense bone. There was an abrupt transition to an inner ground glass or fibro-osseous like zone. This latter material comprised the bulk of the lesion, including the base. The mass was associated with non-destructive expansile remodeling of adjacent osseous structures, including mild expansion of the anterior frontal sinus wall. Corresponding MR images revealed inflammatory-type changes affecting the left frontal calvarium, superior lateral left orbital wall and abutting the lamina papyracea. There was adjacent

Fig. 1 Axial bone window CT image demonstrates a polypoid osseous mass of mixed radiodensity extending into the right and left frontal sinuses. Residual left airspace is opacified

Fig. 2 Coronal CT image of the bony mass reveals a thick outer shell of compact bone around ground glass-appearing material extending to the base

Fig. 3 On the sagittal CT image there is extension into the frontoethmoidal recess with opacification of left anterior ethmoidal air cells

patchy meningeal enhancement along the lateral inferior convexity of the left frontal lobe without intracranial extension.

Fig. 4 In this coronal CT image the mass extends in the posterior portion of the opacified left frontal sinus where there is thinning and loss of integrity of the orbital wall along with opacification of the ethmoid bulla below the osteoma. A focus of compact bone is present at the superior aspect of the mass

Fig. 5 Dense, Paget-like bone at the periphery of the osteoma transitions into areas closely resembling osteoblastoma

Treatment

The mass was removed in a piecemeal fashion via frontal sinus exploration and non-endoscopic Lothrop procedure. There was no evidence of recurrent or residual lesion at the 3 month follow-up.

Diagnosis

Grossly, the specimen consisted of multiple fragments of gray-tan bone measuring 8.0 cm in aggregate. Microscopic examination revealed lobulated portions of dense, sclerotic bone that were partially surfaced by a thin layer of respiratory mucosa. The sclerotic bone blended or

transitioned into areas characterized by irregular trabeculae of immature woven bone rimmed by prominent, enlarged and occasionally confluent osteoblasts (Figs. 5, 6). There were frequent admixed osteoclasts. The intertrabecular fibroblastic tissue was loosely textured and moderately cellular with prominent vascularity and hemorrhage. Paget-like bone was present adjacent to these areas. Additional portions of the lesion were characterized by mature bony trabeculae set in a fibrous background. The clinical, radiographic and morphologic findings were characteristic of a sino-orbital osteoma with osteoblastoma-like features.

Discussion

The sino-orbital osteoma with osteoblastoma-like features is a diagnostically challenging variant of osteoma. Osteomas are benign bone forming lesions which are slow-growing and often composed of densely sclerotic “ivory” bone. They are the most common tumors found within the paranasal sinuses [1]. Osteomas occur across a wide age range from adolescence to older adults with the majority of cases diagnosed during the 4th and 5th decades of life [2, 3]. There appears to be a slight gender predilection, with males accounting for 54–60 % of reported cases [3, 4]. Very rarely, sinonasal osteomas arise as a component of Gardner syndrome and in the current case the patient was otherwise healthy and did not manifest any features of the syndrome [4, 5]. Clinically sino-orbital osteomas can present with a broad spectrum of signs and symptoms that may include pain, headache, facial distortion, proptosis, polyps, rhinorrhea, epistaxis, periorbital edema and visual changes; true mucoceles and infections may result when frontal sinus osteomas cause nasofrontal duct obstruction, whereas pneumatocele, cerebrospinal fluid leakage, meningitis and cerebral abscess can occur from tumors that erode the supraorbital plate and dura mater [4, 6, 7]. These symptoms are due to mass effect and it has been suggested that osteomas remain asymptomatic until they reach a certain size [8]. Sino-orbital osteomas usually consist of a mass of mature compact or trabecular bone which protrudes as a polypoid growth into the affected sinus space or orbit. They most commonly involve the frontal sinus followed in descending order by the ethmoid sinus, maxillary antrum, and sphenoid sinus [2–5]. The orbit may also be a primary site of involvement [4]. Nearly 40 % of sino-orbital osteomas have foci of enlarged osteoblasts and immature bone in a loose fibrovascular stroma that are histologically indistinguishable from osteoblastoma; such tumors have been designated sino-orbital osteomas with osteoblastoma-like features [4].

Fig. 6 Osteoblastoma-like areas demonstrated mineralized and unmineralized trabeculae rimmed by prominent, enlarged osteoblasts with “bridging” osteoblasts joining trabeculae. The interosseous stroma is loosely textured and moderately cellular with prominent vascularity and hemorrhage. Mild cytologic atypia, similar to that seen in conventional osteoblastoma is evident

The radiographic differential diagnosis primarily includes fibrous dysplasia and ossifying fibroma, which are both expansile tumors of mixed radiodensity [9]. Fibrous dysplasia usually shows a homogenous ground glass appearance and ossifying fibroma is characteristically a well-defined mass with a patchy pattern of osteoblastic and osteolytic areas surrounded by a thin sclerotic margin [9]. While true osteoblastoma and osteoid osteomas are also in the differential, osteoid osteomas are exceptionally uncommon in the craniofacial bones [2]. They classically present with a lucent central nidus surrounded by a zone of reactive sclerosis and do not demonstrate polypoid, intra-cavitary growth. Additionally, true osteoblastomas are generally found in the axial skeleton and long bones, but rarely affect the paranasal sinuses. It is likely that a significant portion of published reports of osteoblastomas in the ethmoid and frontal sinuses are, in fact, examples of osteomas with osteoblastoma-like features [4]. CT imaging is better suited than MR images for accurately delineating sino-orbital osteomas with osteoblastoma-like features. With MR imaging, the signal void peripheral zone of the lesion may not be distinguished from air in paranasal sinuses, causing confusion if not evaluated in conjunction with corresponding CT images [9].

The primary histologic differential diagnosis includes osteoblastoma and osteoid osteoma, both of which are characterized by anastomosing trabeculae of osteoid or woven bone rimmed by enlarged osteoblasts and osteoclasts and set in richly vascularized fibromyxoid stroma.

Identical areas may be quite conspicuous in osteomas with osteoblastoma-like features but these lesions also contain much more mature compact and conventional cancellous bone [4]. Microscopically, ossifying fibromas and fibrous dysplasia, are readily separated based on the usual presence of abundant collagenous fibroblastic tissue. The presence of mild cytologic atypia in the large osteoblasts rimming thin trabeculae of osteoid may result in consideration of an osteoblastic osteosarcoma, however evidence of maturation and an absence of atypical stromal cells precludes a diagnosis of osteosarcoma [7]. Ultimately, distinguishing osteoma with osteoblastoma-like features from these entities requires correlating both morphologic and radiographic findings.

Sino-orbital osteomas with osteoblastoma-like features are sufficiently distinctive to warrant awareness, but do not appear to behave differently than other sinonasal osteomas [4]. The osteoblastoma-like areas may represent areas of active bone remodeling, a presumption supported by the frequent presence of Paget-like bone [4]. Care should be taken to avoid misinterpretation as a true osteoblastoma. Surgical options for removal of paranasal sinus osteomas include endoscopic resection, external access or a combination of both [8].

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