Surgical management and outcome of large orbitocranial osteomas

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Object. The goal of this study is to review the surgical management and outcome of patients who were treated for large orbitocranial osteomas at Gulhane Military Medical Academy over a period of 7 years.

Methods. Twenty-one patients with large orbitocranial osteomas were evaluated retrospectively. All patients were male and between 19 and 25 years old. Surgery was performed in all patients. The main surgical procedure was resection of the osteoma using orbitotomy and/or craniotomy followed by orbital reconstruction and cranioplasty. Cranioplasty was performed in 16 patients, using methyl methacrylate in 5 patients (31%) and porous polyethylene in 11 patients (69%). Thin, flexible, porous polyethylene was preferred for orbital reconstruction in 10 patients. The cranioplasty materials were attached to the intact bone using miniplates.

Results. There were no severe postoperative complications. Mild transient periorbital ecchymosis was noted in 19 patients. The mean follow-up period was 11.7 months (range 9–24 months) after surgery. No tumor regrowth was observed in any patient at the end of the follow-up period.

Conclusions. Large osteomas of the orbitocranial region must be resected for cosmetic and functional reasons. Selection and planning of the surgical technique should be based on the direction of the tumor growth and on the size of the tumor and the structures that are compressed by the tumor. (DOI: 10.3171/JNS/2008/109/9/0472)

Keywords: cranioplasty • cranium • orbit • osteoma

Osteomas are the most common of the primary benign bone tumors of the skull and facial structures. They occur in the cranial vault, maxilla, external auditory canal, and the mandible, but most frequently in the paranasal sinuses and mastoid cells. Osteomas are mainly asymptomatic, and account for 0.43% of tumors in the general population. Osteomas are an incidental finding on 1% of plain radiographs of the sinuses, and on 3% of CT scans of the sinuses.

Osteomas of the paranasal sinuses occur mainly in the frontal and ethmoid sinuses; osteomas of the maxillary and sphenoid sinuses are very rarely observed. Frontal sinus osteomas include 57% of all paranasal sinus osteomas. The most common period of presentation of these tumors occurs in patients between the second and fifth decades of life, with a male predominance of approximately 3:1.

Paranasal sinus osteomas have a tendency to grow slowly. Koivunen and colleagues reported a mean growth rate of these tumors of 1.61 mm/year (range 0.44–6 mm/year), and therefore these tumors are generally asymptomatic. The base of these lesions is frequently narrow and the mass is pedunculated. Many of these lesions may be removed via an orbital route. Tumor size, location, and extension determine the clinical manifestations. Most paranasal sinus osteomas have orbital and craniofacial extensions.

Surgical treatment of asymptomatic osteomas may be indicated for cosmetic reasons, but specific conditions are considered as indications for surgery. The treatment generally consists of en bloc resection or curettage of the tumor using a high-speed drill. For large orbitocranial osteomas, combining the craniotomy with an orbitotomy makes a single-stage radical excision possible.

Only a few studies of large orbitocranial osteomas have been published previously. The aim of this study is to retrospectively evaluate patients who had large orbitocranial osteomas and to analyze the clinical, radiological, and surgical aspects of these lesions.

Methods

Patient Population

Between 2000 and 2006, 21 patients with large orbitocranial osteomas were treated surgically in the Department of Neurosurgery. The patient population consisted of young-adult male patients ranging in age from 19 to 25 years (mean 21.7 years) with large craniofacial osteomas involving the paranasal sinuses and presenting with orbital extensions. All of the patients were private soldiers. Headache and a growing mass lesion on the skull were the main complaints. The symptoms and signs of the tumors are summarized in Table 1.
Orbitocranial osteomas

All patients underwent neurological and radiological evaluation in the preoperative period, including: plain radiographs; cranial, orbital, and paranasal sinus CT scans; MR imaging; and sometimes tridimensional cranial CT and scintigraphy (Figs. 1 and 2). The thickness and dimensions of each osteoma were measured. The origin and extension of the tumors were noted (Table 2). Mass lesions larger than 3 cm were considered large tumors. The MR imaging demonstrated the location of the tumor and its relation to nearby structures. Sometimes bone scintigraphy was used for distinguishing osteoma from other skull tumors.

Surgery was performed in all patients. The type of surgery selected was based on tumor size, location, extension, and relationship to underlying brain tissue, dura mater, and orbital structures. Removal of the bone mass lesion via craniectomy, followed by cranioplasty using methyl methacrylate or porous polyethylene and/or orbital reconstruction, was the method used for patients who had a bone tumor growing inward and compressing brain tissue and/or orbital structures. This method was also used in some patients with outgrowing osteomas. Removal of the tumor origin by curettage and high-speed drilling of the sinuses was performed in all patients. A malleable retractor was placed under the orbital roof to prevent the drill or osteotome from damaging the orbital contents. A small diamond bur on a high-speed drill unit was used to remove the osteoma without injury to the underlying structures. The sinuses were successfully cleaned and exenterated of mucosal lining. A small piece of muscle was also placed into the sinuses and was routinely covered with a pericranial flap to isolate the sinuses from the epidural space. Once the osteoma was removed, cranial and/or orbital reconstruction was started. Cranioplasty was the key cosmetic part of the operation and was meticulously performed to avoid cranial asymmetry. Optic nerve decompression was performed in patients who had a narrowed optic canal caused by the osteoma (Table 3).

Postoperative Follow-Up

All patients received antibiotics routinely for at least 72 hours after the operation to prevent skin infection. There was a risk of infection because the paranasal sinuses had been opened. The patients were monitored in the intensive care unit for at least 24 hours, with close monitoring for hemodynamic changes and possible cerebrospinal fluid leaks secondary to dural tears. Medication was administered as needed to provide adequate pain relief.

Computed tomography scans were performed in the early postoperative period as a routine procedure. All patients were followed up with neurological examinations and serial CT scans at 3-month intervals for 6 months, and at 6-month intervals until the end of the 1st year. A CT scan was also performed at the end of the follow-up period in each case.

Results

Twenty-one consecutive young adult patients who had large paranasal sinus osteomas with orbitocranial extensions were treated surgically over a period of 7 years. All patients were males, with a mean age of 21.7 years (range 19–25 years). The mean follow-up period was 11.7 months (range 9–24 months) after surgery. Headache and a slowly enlarging mass were the presenting signs in all patients. Eight patients had normal neurological examination results. The median duration of the preoperative tumor growth period, from the time the osteoma was perceivable by the patient, was 3.6 years (range 2–7.5 years).

The origins of the osteomas were the frontal sinus in 11 patients (52%), ethmoid sinuses in 8 (38%), sphenoid sinus in 1 (5%), and maxillary sinus in 1 (5%). The osteomas in the frontal and ethmoid sinuses extended to the orbital walls and frontal bone together; the osteomas originating at the sphenoid sinus extended to the orbital walls and sphenoid bone; and the osteomas originating in the maxillary sinus extended to the orbital walls and zygoma (Table 2; Fig. 3).

The mean diameter of the osteomas was 5.5 cm (range 4–9 cm). Optic nerve compression was observed in 6 patients, and the orbit and frontal lobe were compressed by the tumor in 11 patients.

A craniectomy and removal of the orbital wall, including the tumor and overlying bone tissue, followed by cranioplasty were performed in 16 patients (76%; Table 3). A cranioplasty was performed using methyl methacrylate in 5 (31%) of the 16 patients and porous polyethylene in 11 patients (69%). For orbital reconstruction in 10 patients, using the thin, flexible, porous polyethylene was preferred. The cranioplasty materials were attached to the intact bone using miniplates. The paranasal sinuses were scraped with a curette and drilled using a high-speed drill for tumor removal in all patients. Optic canal decompression was performed in 6 patients.

The histopathological examination of the tumors confirmed the diagnosis of osteoid osteoma of the paranasal sinuses. There were no severe complications in the early postoperative period such as hemorrhage, ocular disturbance, loss of vision, or cerebrospinal fluid fistulas. Mild postoperative peri orbital ecchymosis was noted in 19 patients. In 1 patient, the cranioplastic material, formed

**TABLE 1**

<table>
<thead>
<tr>
<th>Symptom/Sign</th>
<th>No. of Patients</th>
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<tbody>
<tr>
<td>headache</td>
<td>21</td>
</tr>
<tr>
<td>slowly growing peri orbital mass</td>
<td>21</td>
</tr>
<tr>
<td>orbital pain</td>
<td>12</td>
</tr>
<tr>
<td>proptosis</td>
<td>11</td>
</tr>
<tr>
<td>eye movement disturbance</td>
<td>8</td>
</tr>
<tr>
<td>normal neurological examination results</td>
<td>8</td>
</tr>
<tr>
<td>diplopia</td>
<td>7</td>
</tr>
<tr>
<td>decrease of visual acuity</td>
<td>5</td>
</tr>
<tr>
<td>anosmia or olfaction disturbance</td>
<td>5</td>
</tr>
<tr>
<td>dacryocystitis</td>
<td>3</td>
</tr>
<tr>
<td>seizure</td>
<td>1</td>
</tr>
</tbody>
</table>

**Operative and Imaging Procedures**

Postoperative Follow-Up
with methyl methacrylate, was removed because of infection 6 months after the operation. We detected the movement of the porous polyethylene cranioplastic material, which was attached to the intact bone using miniplates, at the 1st year follow-up in another patient; that patient underwent another operation and the material was fixed again (Table 4).

None of the patients had complaints, and the physical examination results were normal in all patients at the end of the follow-up period. No recurrence or residual osseous tumor was observed on plain radiographs and CT scans in any patient during this period.

**Discussion**

Orbital osteomas were first described in 1506 by Veigra,4 who reported the case of an osteoma of the superior medial angle of the orbit diagnosed in a young woman who presented with an exophthalmus with facial distortion. An osteoid osteoma is a benign osteoblastic lesion and constitutes 1% of all bone tumors and 11% of benign bone lesions.8

An orbital location of osteomas is particularly rare, and their incidence represents 0.9–5.1% of all orbital tumors.22,23 The mean age of patients with orbital osteomas is 46.4 years, with a male to female ratio of 1.85. This ratio is believed to be secondary to the fact that men are exposed to more traumas than women and also have larger sinuses. In our patient series, all patients were young adult males, with a mean age of 21.7 years, and all were private soldiers.

The osteoma is a mesenchymal tumor that develops at the expense of bones and sinuses of the face. The causes of osteoma are controversial; the most accepted theories are genetic, traumatic, embryological, and infectious causes.10,16,17,19,22,26 According to the genetic theory, genetic factors probably play an important role in the development of osteomas included in Gardner syndrome, characterized by intestinal polyps and multiple osteomas.28 Theories of traumatic and embryological causes explain the metaplasia of the connective tissue and the sinusal mucosa behaving like a periost and developing a bony substance. The infectious theory has been widely discussed, according to which bone metaplasia is secondary to a chronic infectious and inflammatory state.3 In favor of the developmental theory is the fact that many

**Fig. 1.** Images obtained in a 21-year-old-male patient with a large left orbitofrontal osteoma originating from the left frontal sinus. A: A coronal T1-weighted MR image shows a mass lesion originating from the floor of the left orbit, extending to the superior, inferior, and left aspects of the frontal lobe. The structures of the left orbit are displaced inferiorly and anteriorly. B: Coronal CT scan reveals that the lesion is isodense with the bone. C and D: The anteroposterior (C) and lateral (D) radiographs demonstrate that the lesion is likely an osteoma.
Orbitocranial osteomas appear to arise at the junction of the ethmoid and frontal sinuses, a location in which membranous and cartilaginous tissues meet during embryonic development. Trauma during puberty, when bone development is at its maximum, has been implicated in several cases of osteomas. Sinusitis may stimulate osteoblastic proliferation within the mucoperiosteal lining of the sinuses, causing tumor formation. In our patient series, we did not find any history of trauma or family history of osteoma. Therefore, we could not determine the real causes of the lesions in our cases.

The growth rate of an osteoma is very slow, from 12 to 30 years according to reported series, even though after incomplete excision, relapse may occur after 2–8 years. The median duration of the preoperative tumor growth period in our cases was 3.6 years.

Symptoms in patients with large orbitocranial osteomas are generally of late onset and are a consequence of tumor growth and compression of neighboring organs. Headache and facial pain localized over the area of the osteoma are the most common presenting symptoms; proptosis is the major ophthalmological sign. Other possible orbital symptoms include diplopia secondary to oculomotor muscle involvement, choroidal folds, papil-

**TABLE 2**

<table>
<thead>
<tr>
<th>No. of Patients</th>
<th>Tumor Origin</th>
<th>Tumor Extension</th>
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<tbody>
<tr>
<td>11</td>
<td>frontal sinus</td>
<td>orbital roof, superoposterior orbital wall, frontal bone</td>
</tr>
<tr>
<td>8</td>
<td>ethmoid sinus</td>
<td>inferior and medial orbital wall, frontal sinus, medial part of the frontal bone</td>
</tr>
<tr>
<td>1</td>
<td>sphenoid sinus</td>
<td>posteroinferior, medial, and inferomedial orbital wall; sphenoid bone</td>
</tr>
<tr>
<td>1</td>
<td>maxillary sinus</td>
<td>inferior and lateral orbital wall, zygoma</td>
</tr>
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lary edema, optic nerve compression and atrophy, orbital cellulitis, and lacrimal tract obstruction. The extraocular manifestations may include facial deformity, rhinorrhea, and anosmia, as well as psychological disturbances such as altered mental status, confusion, amnesia, and others. Headache and a slowly growing periorbital mass lesion were the most common complaints of patients in our series, and proptosis was determined in 11 cases.

Management of paranasal sinus osteoma with orbitocranial extension remains controversial. Resection is advised when these lesions are symptomatic or when they exhibit rapid growth.20 Although asymptomatic and small orbital osteomas are generally left untreated and require a yearly clinical and radiological follow-up evaluation, the progressive tumor growth may seriously affect vision by compressing the optic nerve or the orbital vascular system, eventually destroying the optic bulb completely. 2,4 Therefore, in cases of orbital involvement, it is always necessary to resort to surgical removal of the lesion. Surgical treatment is particularly indicated in cases of a voluminous osteoma, and is complicated in sphenoidal localization threatening visual function, even though the tumor is small and asymptomatic. Surgery is indicated more rarely for aesthetic worries.4 In our case series, only 8 of 21 patients had normal neurological examination results; the other patients had some neurological and ophthalmological clinical evidence of the tumor.

Treatment of osteomas generally consists of en bloc resection or curettage.10,14 Traditionally, resection has been performed by means of a variety of open approaches, such as frontobasal craniotomy, coronal approaches, or mixed approaches (transfacial and neurosurgical); use of an orbitotomy is usually reserved for osteomas with orbitocranial extensions.23 We generally performed the resection of the tumor followed by cranioplasty. We reconstructed the bone defect using methyl methacrylate or porous polyethylene. Especially for orbital reconstruction, the thin, flexible, porous polyethylene was used for obtaining good cosmetic results. Paranasal sinuses were scraped with a curette and drilled using a high-speed drill for removing the tumor in all patients. If open surgery carries severe risks, endoscopic resection could be per-
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formed safely. But this approach is not possible for voluminous orbitocranial osteomas.

Conclusions

Osteomas of the paranasal sinuses are usually asymptomatic. If they become voluminous, they may cause orbital and cranial manifestations and serious complications. The large paranasal osteomas with orbitocranial extension must be removed for cosmetic and functional reasons. Selection and planning of the surgical technique should be based on the direction of the tumor growth and on the size of the tumor and the structures that are compressed by it.

Disclaimer

The authors do not report any conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

References


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