

CASE REPORT

A case of invasive meningioma involving the maxillary sinus

Maksiller sinüs tutulumu gösteren invaziv meningiom

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Meningiomas account for nearly 15% of primary brain tumors, but extracranial meningiomas are very rare. We presented a case of invasive maxillary sinus meningioma. A 50-year-old man presented with facial tenderness and severe pain in the left cheek. He had a prior surgery for a meningioma in the left frontal lobe eight months before. Physical examination and computed tomography showed a mass in the left maxillary sinus. Histopathological result of the biopsy obtained via the Caldwell-Luc approach was invasive meningioma. The mass was removed with the sinus mucosa. The histology of the resected specimen was compatible with invasive angioblastic meningioma. Postoperative radiotherapy was administered because of residual intracranial tumor. No recurrence was detected over an 11-month follow-up period.

Key Words: Maxillary sinus neoplasms/pathology/surgery; meningioma/pathology/surgery.

Meningiolar primer beyin tümörlerinin yaklaşık %15'ini oluşturmalarına karşın, ekstrakraniyal tutulum çok nadirdir. Bu yazıda invaziv maksiller sinüs meningiomlu bir olgu sunuldu. Elli yaşında bir erkek hasta sol yanak bölgesinde şiddetli ağrı ve hassasiyet şikayeti ile kliniğimize başvurdu. Öyküsünde, sol frontal lobda yerleşik bir meningiom için sekiz ay önce geçirilmiş bir ameliyat vardı. Fizik muayene ve bilgisayarlı tomografi ile sol maksiller sinüste bir kitle saptandı. Caldwell-Luc yöntemiyle alınan biyopsinin histopatolojik sonucu invaziv meningiom olarak bildirildi. Kitle sinüs mukozasıyla birlikte çıkarıldı. Cerrahi örneğin histopatolojik tanısı invaziv anjiyoplastik meningiom idi. Hastaya, rezidüel intrakraniyal tümör nedeniyle ameliyat sonrasında radyoterapi uygulandı. On bir aylık takip süresi içinde nüks görülmedi.

Anahtar Sözcükler: Maksiller sinüs neoplazileri/patoloji/cerrahi; meningiom/patoloji/cerrahi.

Meningiomas originate from arachnoidal cap cells forming the outer lining of the arachnoid membrane. They account for about 15% of primary brain tumors and 25% of spinal cord tumors.^[1] Extracranial meningiomas rarely occur in the orbit, calvarium, neck, temporal bone, paranasal sinuses, and mandible.^[1-3]

We presented a rare case of meningioma of the maxillary sinus.

CASE REPORT

A 50-year-old man presented with complaints of severe pain and slight swelling in the left cheek that had progressed slowly during the past month. He had a prior surgery for a meningioma in the left frontal lobe eight months before. Otolaryngological examination showed a scar tissue in the left frontal region and a slight swelling in the left fossa canina

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(Fig. 1). He complained of facial tenderness and severe pain on palpation of the maxilla. Neurological examination was normal except for a slight speech disorder that developed after prior neurosurgical operation. Endoscopic evaluation of the nasal cavity and nasopharynx was normal. Computed tomography (CT) showed a mass in the left maxillary sinus with moderate enhancement and a left frontal bone defect. The mass extended posteriorly to the orbit and superior orbital fissure, and to the maxillary sinus (Fig. 2). Upon consultation with the neurosurgery department, a craniofacial resection was not recommended because visual functions were normal. Therefore, a biopsy of the mass was obtained under general anesthesia via the Caldwell-Luc approach. The anterior wall of the maxillary sinus was thinned due to compression of the mass. A large bone window was opened preserving the infraorbital nerve, and a gray, encapsulated solitary mass was seen filling the sinus (Fig. 3a). The mass was removed with the sinus mucosa (Fig. 3b). A bone defect was identified on the posterosuperior wall of the sinus. The sinus cavity was packed with a tampon for three days. Histopathologic examination of the specimen showed uniform round spindle-shaped atypical nuclei arranged in spiral rows, rosettes, and a pale wavy eosinophilic cytoplasm with invisible histologic borders. Moderate mitotic figures were observed (Fig. 4). In immunohistochemical staining, the cytoplasm of the tumor cells was positive for vimentin and cytoplasmic membrane was positive for EMA (epithelial membrane antigen). Desmin, alpha-smooth muscle actin, S-100 protein, and cytokeratin were negative. The histology of the resected specimen was compatible with invasive

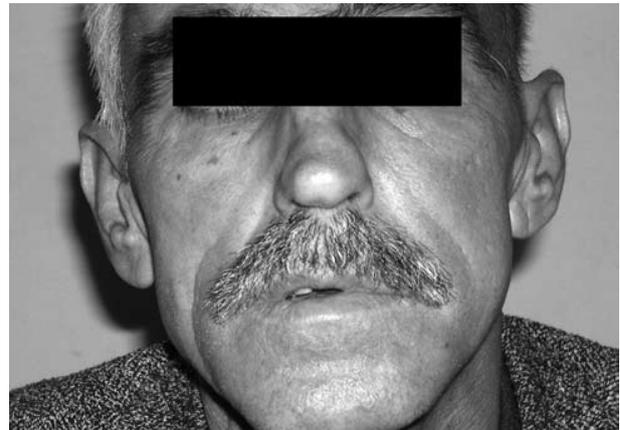


Fig. 1. There was a scar tissue in the left frontal region and a slight swelling in the left fossa canina.

angioblastic meningioma. Radiotherapy was planned because of residual intracranial tumor. No recurrence was detected over an 11-month follow-up period.

DISCUSSION

Meningiomas are seen in adults, with an age range of 20 to 60 years. The peak incidence is around the age of 40. A female preponderance, especially in the spinal cord meningiomas, is very high.^[4,5] Although meningiomas can potentially occur at any site in the meninges, certain intracranial locations are more common including parasagittal and falx meningiomas, and those occurring over the cerebral convexities, in the olfactory groove, on the tuberculum sellae, along the sphenoid ridge, in the cerebellopontine angle, along the clivus, and at the foramen magnum. In some cases (20%) combination of intracranial and extracranial sites can be seen.^[4,5]

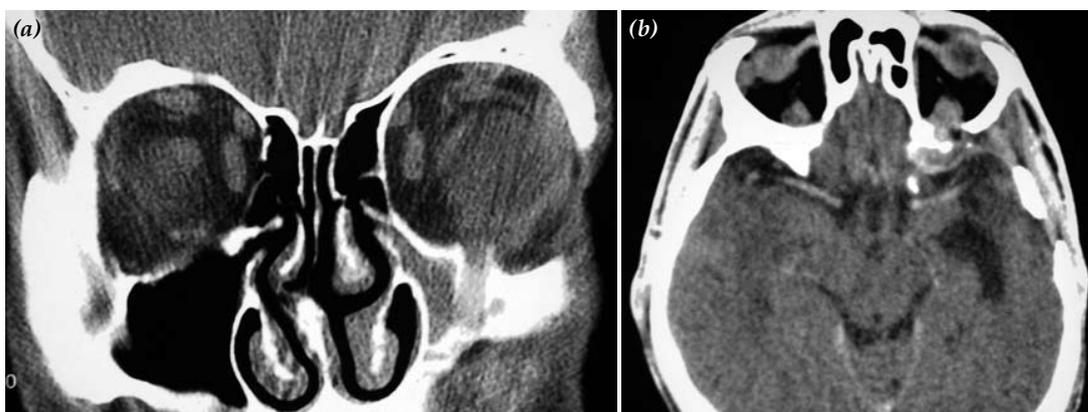


Fig. 2. (a) Coronal and (b) axial sections of computed tomography showing a mass in the left maxillary sinus with moderate enhancement and a left frontal bone defect. The mass extended posteriorly to the orbit and superior orbital fissure, and to the frontal lobe.

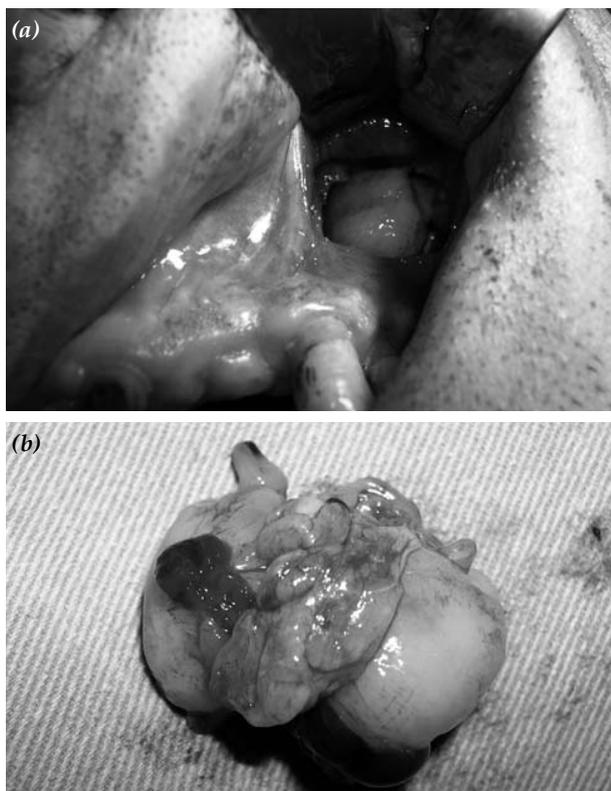


Fig. 3. (a) A gray encapsulated solitary mass filling the sinus. (b) The mass and mucosa of the maxillary sinus.

Meningiomas are considered to originate from arachnoid cells. These cells are found both along the intracranial sinuses and the dural investment of the cranial nerve foramina. They may be present in the temporal bone extensions of dura, and may be seen as far as the greater and lesser petrosal nerves.^[6] Although all true extracranial meningiomas probably arise from ectopic arachnoid lining cells, their presentation and location suggest at

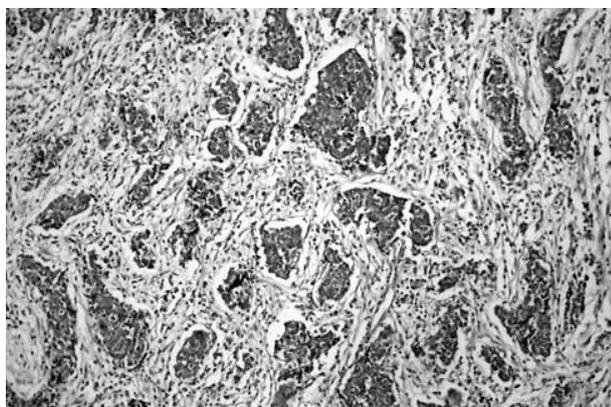


Fig. 4. Moderate mitotic figures (H-E x 4).

least two different pathogenetic mechanisms. One form of extracranial meningioma termed type-1 by Lopez et al.^[7] occurs in children and young adults and is usually present at birth. The lesions are situated in the skin of the scalp, forehead, and paravertebral region and as a result, may be mistaken clinically for cutaneous lesions, including epidermal inclusion cysts, skin tags, and *nervi*.^[7] The second form of extracranial meningioma, type-2, may occur at any age, but is usually seen in adults. These tumors are situated in the vicinity of the sensory organs or along the paths of the cranial and spinal nerves.^[7] Symptoms associated with the tumor are related to its size, location, and growth rate. Histologically, these lesions are indistinguishable from the ordinary intracranial meningioma.^[1] This may explain many intracranial and pericranial meningioma cases. According to another opinion, primary extracranial meningiomas are in intimate association with cranial nerves and are derived from ectopic arachnoid tissue present along the course of the nerves.^[7,8] Many classifications of meningiomas have been made related to their dominant cellular morphology, which may present as meningothelial (syncytial), transitional, fibroblastic, angioblastic, and hemangiopericytoma-like features. The transitional and meningothelial subtypes are reported to be the most common forms.^[9,10] In our case, histopathologic diagnosis was made as angioblastic meningioma. Although angioblastic meningiomas are considered to metastasize more commonly than other varieties, we did not observe any distant metastasis in our case. There is no specific radiological technique for the diagnosis of these lesions. Meningiomas demonstrate good enhancement on CT scans. However, it has been indicated that CT scanning is not specific for extracranial meningiomas.^[11] In our case, CT showed a contrast-enhancing lesion in the left maxillary sinus, extending from the frontal lobe to the maxillary sinus via the superior orbital fissure (Fig. 2).

Surgical excision is the main treatment for meningiomas. Localization of the tumor and the possibility of total excision positively affect the prognosis of extracranial meningiomas. As in our case, radiotherapy can be combined with incomplete resection for palliation.

In conclusion, extracranial meningioma should be suspected in patients presenting with a swell-

ing in the maxillary region. Surgical treatment of extracranial meningioma may not be aggressive because no recurrence has been reported. Invasive maxillary sinus meningioma should be considered in the differential diagnosis of maxillary sinus lesions in cases with tenderness and swelling and a history of cranial meningioma.

REFERENCES

1. Lantos PL, Vandenberg SR, Kleihues P. Tumors of the nervous system. In: Graham DI, Lantos PL, editors. *Greenfield's neuropathology*. 6th ed. New York, NY: Oxford University Press; 1997. p. 727-44.
2. Jones AC, Freedman PD. Primary extracranial meningioma of the mandible: A report of 2 cases and a review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001;91:338-41.
3. Günhan Ö, Karcı B. Sinüslerin benign tümörleri. In: Günhan Ö, Karcı B, editors. *Burun ve sinüs tümörleri*. İzmir: Özen Ofset Ltd. Şti.; 1999. p. 58-67.
4. Nager GT, Heroy J, Hoeplinger M. Meningiomas invading the temporal bone with extension to the neck. *Am J Otolaryngol* 1983;4:297-324.
5. Rietz DR, Ford CN, Kurtycz DF, Brandenburg JH, Hafez GR. Significance of apparent intratympanic meningiomas. *Laryngoscope* 1983;93:1397-404.
6. Nager GT. Meningiomas involving the temporal bone: clinical and pathological aspects. *Ir J Med Sci* 1966;6:69-96.
7. Lopez DA, Silvers DN, Helwig EB. Cutaneous meningiomas-a clinicopathologic study. *Cancer* 1974;34:728-44.
8. Granich MS, Pilch BZ, Goodman ML. Meningiomas presenting in the paranasal sinuses and temporal bone. *Head Neck Surg* 1983;5:319-28.
9. Kershisnik M, Callender DL, Batsakis JG. Extracranial, extraspinal meningiomas of the head and neck. *Ann Otol Rhinol Laryngol* 1993;102:967-70.
10. Batsakis JG. Pathology consultation. Extracranial meningiomas. *Ann Otol Rhinol Laryngol* 1984;93:282-3.
11. Vassilouthis J, Ambrose J. Computerized tomography scanning appearances of intracranial meningiomas. An attempt to predict the histological features. *J Neurosurg* 1979;50:320-7.