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CASE REPORT

Case of primary extracranial meningioma of the maxillary sinus presenting as buccal swelling associated with headache: A case report

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Abstract

BACKGROUND

Meningiomas are benign tumors that originate from the meningothelial arachnoid cells, but they rarely develop extracranially. There is no specific surgical guideline for resecting them in the maxillary sinus, and little is known about their biological behavior and operative management.

CASE SUMMARY

We present a 54-year-old female patient referred to our department with a primary extracranial meningioma that presented as buccal swelling associated with headache. On clinical examination the mass was non-tender, fixed, sessile and non-pulsatile situating in the right maxillary sinus. Computed tomography scan showed a well-defined mass of 7 cm × 6 cm × 6 cm compressing the surrounding structures. Magnetic resonance imaging revealed a well circumscribed heterogenous lesion with necrotic center and relatively hypointense on T2-weighted imaging. Imaging studies revealed no evidence of intracranial extension and metastatic nests. Biopsy showed grade I primary extracranial with low mitotic activity. Total maxillectomy with excision of tumor and adjacent paranasal structures following reconstruction of the orbit and maxilla with tissue patch was done by the maxillofacial surgeon. The biopsy reported fibrous meningioma based on the hematoxylin and eosin section. On immunohistochemistry the tumor cells were positive for vimentin, focally positive for epithelial membrane antigen and CD99 and negative for signal transducer and activator of transcription 6. The mass was removed surgically with reconstruction, and the pathological studies confirmed the diagnosis to be an extracranial meningioma. The present study briefly reviews the current knowledge concerning the diagnosis and treatment of extracranial meningiomas in the head and neck area and offers suggestions for managing extracranial meningiomas in the paranasal sinuses.

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CONCLUSION
To conclude, extracranial meningiomas in the paranasal sinuses may be successfully managed by surgical treatment without evident post-surgery complications.

Key Words: Primary extracranial meningioma; Maxillary sinus; Diagnosis; Surgical treatment; Buccal swelling; Case report

Core Tip: Meningiomas are rare benign tumors originating from the meningothelial arachnoid cells that rarely occur in an extracranial location. So herein we present a rare case of extracranial meningioma in the maxillary sinus of a 54-year-old female presenting with right buccal swelling. Headache was the only symptom. This case mainly focuses on the diagnosis and surgical management with reduced post-surgical complication and provides an insight and surgical guideline in treatment planning.

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INTRODUCTION

Meningiomas are one of the largest groups of brain tumors. They come in two forms: intracranial and extracranial. The extracranial location is very rare. Approximately 6%-17% of all meningiomas can be found in extracranial regions[1]. Male patients are more likely to have extracranial meningiomas[2]. We describe a rare case of primary extracranial meningioma of the maxillary sinus in a 54-year-old female patient presenting as buccal swelling and headache. Regardless of the grade, the recommended treatment is complete surgical excision if possible; we used a combined surgical approach to achieve complete excision of the lesion. The clinical, histological and immunohistochemical features are described. The possible histogenesis and the differential diagnosis are also discussed. Subsequently, we reviewed the literature on this respect.

CASE PRESENTATION

Chief complaints

A 54-year-old female patient presented with right buccal swelling for 2 years and headache for 2 mo to the Department of Head and Neck Oncology Surgery, West China College of Stomatology, Sichuan University.

History of present illness

The patient visited a local hospital and started anti-inflammatory and analgesic drugs as they considered the symptoms to be caused by cold and toothache, but the pain did not improve significantly. The patient again visited Guangyuan People's Hospital seeking further treatment. The biopsy taken showed the spindle cell tumor in the right maxillary sinus, which was further examined by immunohistochemistry. The patient denied any shortness of breath, nausea, dysphagia, hoarseness, loss of consciousness and any neurological or constitutional symptoms at any time.

History of past illness

The patient had no previous medical history.

Personal and family history

She was a non-smoker with no specific family history.

Physical examination

On extra-oral examination the mass located in the right face was non-tender, fixed and non-pulsatile and sessile (Figure 1A). The patient did not have any palpable lymph nodes or associated neck masses. Upon intra-oral examination, an obvious buccal swelling covered with slightly red oral mucosa was present. The majority of the mass was located in the right maxillary sinus and involved the base of the maxilla.

Laboratory examinations

The patient underwent biopsy of the mass using gingival incision extending as far as the upper first molar teeth under local anesthesia. Biopsy reported a grade I primary extracranial meningioma with low mitotic activity. Hematological examinations were within normal limits.

Imaging examinations

Magnetic resonance imaging revealed a well-circumscribed heterogenous lesion with a necrotic center and was relatively hypointense on T2-weighted imaging (Figure 1B and C).

Computed tomography (CT) demonstrated the presence of a large, well-defined soft tissue mass measuring about 7 cm × 6 cm × 6 cm occupying the entirety of the right maxillary sinus, affecting nearby sphenoid and ethmoid sinuses, without affecting the dura mater or endocrinal structures. The surrounding structures were compressed by the mass, and the mass extended from the roof of the oral cavity into the skull base. CT on bone window setting showed an expansive mass with a high density area in the right maxillary sinus. The anterior and lateral walls of the maxillary sinus were thinned and destructed by the expanding mass, with erosion of the wall of the right maxillary sinus as well as orbital floor. (Figure 1D and E). Imaging study based on comprehensive detection of the lesion revealed that there was no evidence of intracranial extension and metastatic nests.

FINAL DIAGNOSIS

Right maxillary meningioma.

TREATMENT

As intracranial invasive meningioma was excluded, the surgery was decided to be performed by the Oral and Maxillofacial Surgeons. Total maxillectomy together with the excision of the tumor and the adjacent paranasal structures, following reconstruction of the orbit and maxilla with tissue patch was performed.

On June 6, 2016, under general anesthesia "extended resection of right maxillary meningioma; right maxillary extended resection; inferior turbinate partial resection; middle turbinate partial resection; right-sided canal neurotomy; right trigeminal peripheral branch transection; A1 extraction; A1-A7 gingival flap; and free skin patch repair" was performed.

Frozen pathology showed spindle cell tumor with extensive necrosis in the right maxilla, which was confirmed by extensive biopsy and immunohistochemical staining. The operation lasted for 2 h and 5 min. The blood loss was 650 mL, and the fluid infused was 2600 mL during the operation. After the operation, the patient returned to the intensive care unit. The vital signs of the patient were observed. Ceftriaxone 2.0g IV BD for 4 d was used to treat infection and prevent intracranial infection. Postoperative nutritional support and antitumor therapy were used.

The entire tumor specimen was submitted for histology, and fresh tissue was fixed in formaldehyde solution for ultrastructural analysis. Histologically, the specimen consisted of epithelioid lobulated tissue, separated by abundant collagen fibers (Figure 2A and B). Image analysis at high magnification showed a thick fibrous capsule and was composed of interwoven fascicles of spindle-shaped meningiocytes and collagen fibers that were arranged into lobules. The tumor cells had abundant cytoplasm and indistinct cytoplasmic borders, arranged in whorled and lobulated patterns. There was osteoid formation in the tumor. No cytologic atypia or necrosis

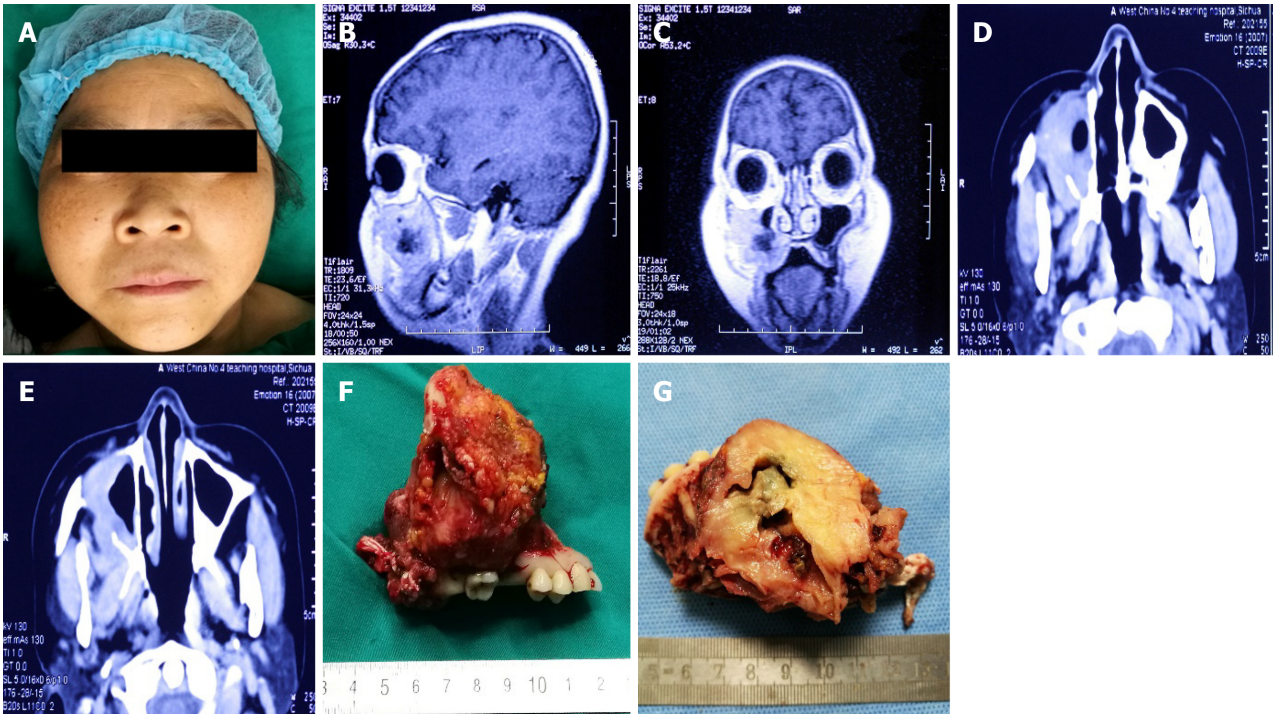

Figure 1 Characterization of imaging studies and gross finding. A: Facial swelling measured about 4 cm in diameter on right side; B: Magnetic resonance imaging (sagittal section) demonstrated a soft tissue mass with a necrotic center compressing adjacent structures; C: Magnetic resonance imaging (coronal view) demonstrated a soft tissue mass with a necrotic center compressing the right maxilla; D and E: Computed tomography imaging demonstrated a soft tissue mass with a necrotic center compressing adjacent structures, red arrow showing the mass compressing the anterior wall of the right maxilla; F: The mass appeared to be lobulated and yellow-white measuring about 8 cm in diameter; G: On hemisection, the mass showed a well-circumscribed heterogenous lesion with a necrotic center.

were discovered, but some mitoses were present. The specimen showed abundant cytoplasm and indistinct cytoplasmic borders, arranged in whorled and lobulated patterns (Figure 2C and D). Based on the hematoxylin and eosin sections, the lesion was diagnosed as a fibrous meningioma.

Immunohistochemically, the tumor cells were strongly positive for vimentin (Figure 3A), focally positive for epithelial membrane antigen (Figure 3B) and CD99 (Figure 3C). The cells showed negative staining for signal transducer and activator of transcription 6 (Figure 3D) and CD34 (Figure 3E). The MIB-1 (Ki-67) labeling index was 15% (Figure 3F), *i.e.* focally positive.

OUTCOME AND FOLLOW-UP

The patient was discharged with the following advice: perform mouth opening exercises; fabricate lumbar appendage in Prosthodontic Department a month later; radiotherapy should be done; proper nutritional support, proper oral hygiene and proper wound care; avoid spicy, acidic and irritating foods; and review after discharge for 1 mo and follow-up for discomfort.

DISCUSSION

Meningiomas can exist as intracranial or extracranial brain tumors and are benign, slow-growing tumors. The extracranial location accounts for 2% of all these tumors[3] and found most often in male patients and in young individuals[4]. Due to their unusual symptoms and lack of prevalence, primary extracranial meningiomas are often misdiagnosed[5]. Fortunately, 80% of extracranial tumors are benign[6]. Cases of extracranial meningioma of the sinonasal tract[7], retromolar area[8], eyebrows[9], pelvis[10], *etc.* have also been reported. Some of the published reports of extracranial meningiomas are listed in Table 1. Histologically, primary extracranial meningiomas do not differ from intracranial, and most of these tumors are sporadic with unclear

Table 1 Published case reports of primary extracranial meningioma

Ref.	Year of publication	Site of primary extracranial meningioma	Diagnostic tests	Histology	Treatment performed
Maharjan *et al*[15]	2018	Nasal cavity	Contrast-enhanced CT of the nose and paranasal sinuses	WHO grade II atypical transitional meningioma	Endoscopic excision of the mass
Kim *et al* [18]	2018	Forehead	CT scan	Lobular architecture composed of tumor cells with eosinophilic cytoplasm and indistinct cell border	Excisional biopsy under local anesthesia
El-Daly *et al* [1]	1997	Maxillary antrum	CT scan	Interlacing bundles of bland-appearing spindle cells associated with calcific deposit	Medial maxillectomy with complete removal of the tumor
Ho *et al*[5]	1980	Right nasal cavity	Sinus x-ray and CT	Clearly demarcated meningioma with fibrous capsule and well-preserved pseudostratified respiratory epithelium	Ablation of the right frontal sinus, external ethmoidectomy and excision of the right middle turbinate
Nur *et al*[12]	2006	Right pelvic cavity	Pelvic sonogram	Lobulated pattern composed of solid sheets of tumor cells separated by connective tissue septae	Exploratory laparotomy with optimal debulking of the pelvic tumor
Albsoul *et al* [11]	2015	Right side neck mass	CT and MRI	Meningothelial cells with intranuclear inclusion and multiple psammoma bodies	Partial excision of the mass
Takeshima *et al*[9]	2004	Right ovary	Abdominal CT	Mature cerebral tissue was also noted. Melanocytes with black pigment were scattered in the peripheral region of the brain tissue	Right salpingo-oophorectomy
Lingen *et al* [8]	1995	Right maxillary sinus	CT	Bundles of ovoid and spindle-shaped cells arranged in broad bands	Total maxillectomy
Rege *et al* [16]	2017	Right retromolar area	CBCT	Spindle cell neoplasm, without evidence of atypia, whorls suggesting meningothelial origin	Partial resection of the mandible and reconstruction with autogenous iliac tricortical bone
Lee *et al*[17]	2017	Left eyebrow	CT	Tumor cells arranged in sheets or whorls, with occasional psammomabodies	Surgical excision
This Study	(Present case)	Maxillary sinus	CT and MRI	Epithelioid lobulated tissue, separated by abundant collagen fibers	Total maxillectomy with excision of tumor

MRI: Magnetic resonance imaging; CT: Computed tomography; WHO: World Health Organization; CBCT: Cone-beam computed tomography.

etiology[11]. Primary extracranial meningiomas have been considered as arising independently from cranial nerve sheaths or from extracranial embryonic rests of arachnoid cells and as extracranial metastases of a primary intracranial meningioma, but their origin has not been completely established[12].

The present case shows the clinical and imaging aspects of extracranial meningioma of the maxillary sinus in an elderly lady. Primary extracranial meningioma of the paranasal sinuses is rare[13]. In general, the most common signs and symptoms of paranasal sinus meningiomas may mimic cases of sinusitis with nasal obstruction, anosmia, facial pressure or pain, epistaxis and rhinorrhea[14,15]. Meningiomas in the extracranial space often present with nonspecific symptoms until the tumor has reached a significant size. This was the case with our patient who had buccal swelling for 2 years, which has been neglected by the patient until the headache started. Clinical examination should be comprehensive because more than 10% of cases may remain asymptomatic even in advanced stages[15]. Imaging studies, especially CT and magnetic resonance imaging scans, have proved to be useful in the diagnosis and management of meningiomas. The differential diagnosis should include a variety of benign and malignant neoplasms such as melanoma, olfactory neuroblastoma, carcinoma, hemangioma, sarcoma and aggressive psammomatoid ossifying fibroma [10,14]. Histology is therefore essential, and the general histologic features and immunohistochemically findings can usually differentiate between these tumors, as extracranial meningioma presents with solid nests of meningothelial cells arranged in sheets or whorls with a fibroadipose background[5,13]. Immunohistochemistry is helpful in confirming the diagnosis; extracranial meningiomas tend to show strong positivity towards vimentin and epithelial membrane antigen, as indeed occurred in

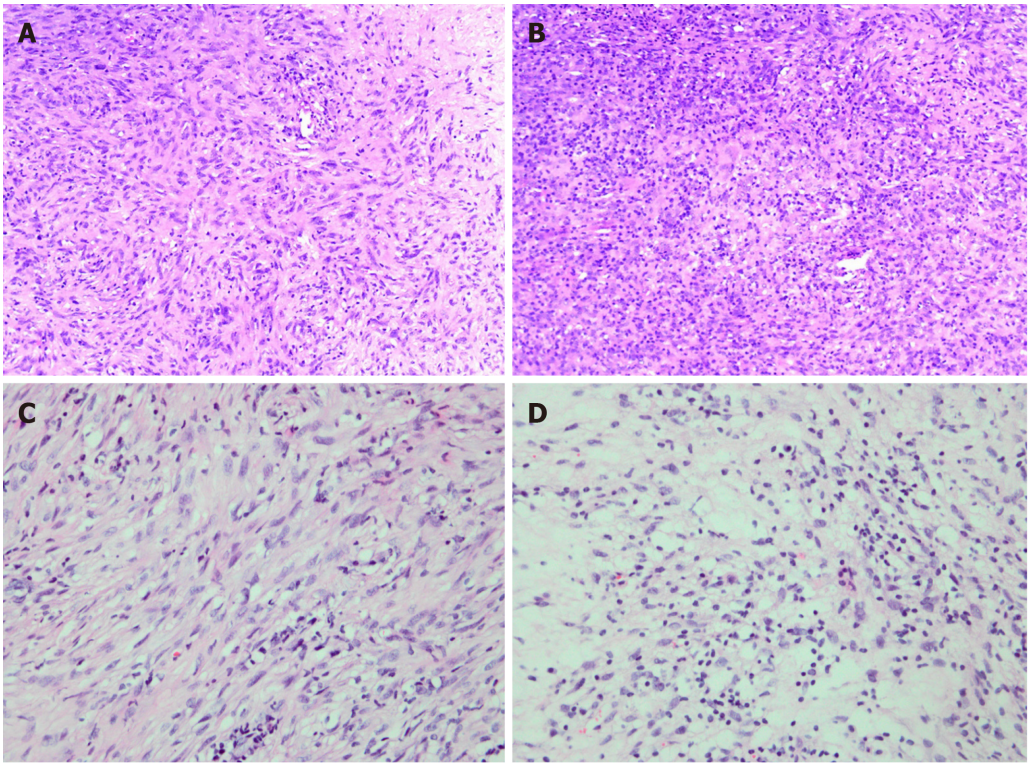

Figure 2 Histological features of extracranial meningioma. A and B: The specimen showed epithelioid lobulated tissue, separated by abundant collagen fibers (hematoxylin and eosin: × 100, × 40); C and D: The specimen showed abundant cytoplasm and indistinct cytoplasmic borders, arranged in whorled and lobulated patterns (hematoxylin and eosin: × 100, × 40).

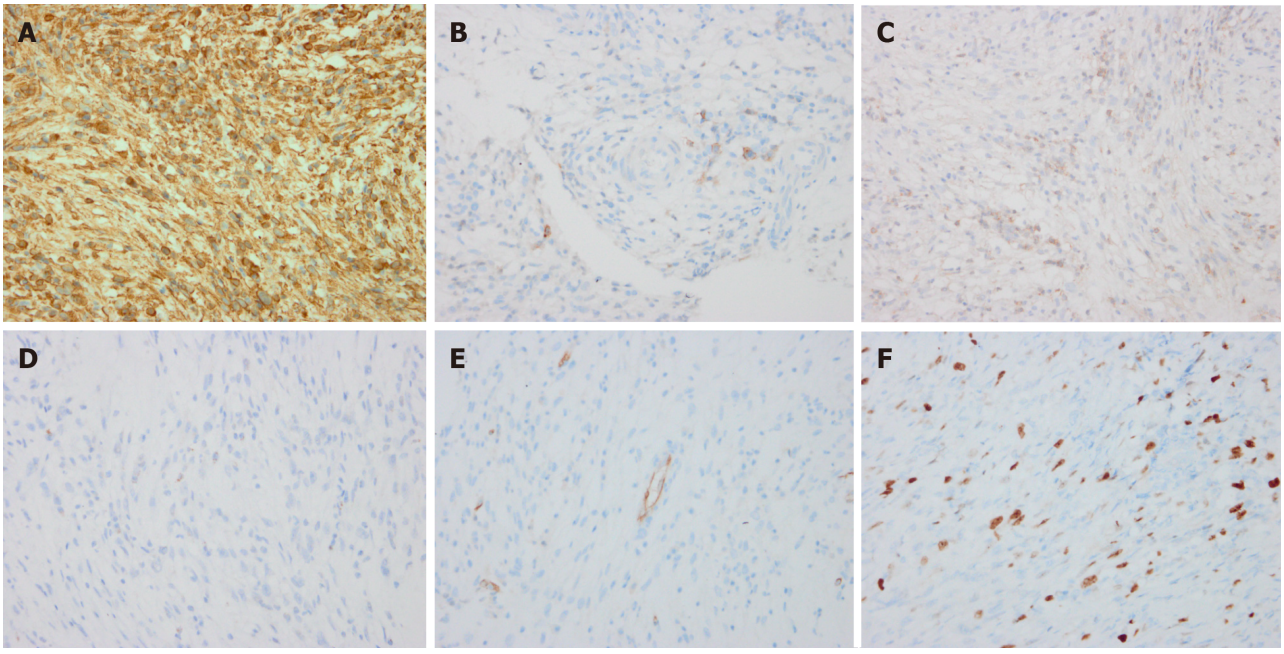

Figure 3 Immunohistochemical findings of the lesion. A: The tumor cells were strongly positive for vimentin; B: The tumor cells were focally positive for epithelial membrane antigen; C: The tumor cells were focally positive for CD99; D: The tumor cells were negative for STAT6; E: The tumor cells were negative for CD34; F: The tumor cells were focally positive for Ki-67.

our patient, and are focally positive for CD99 and Ki-67.

Both CT and magnetic resonance imaging are essential in preoperative surgical planning. Surgery is the only curative treatment, and surgical excision of the mass should be performed if possible. External beam radiation therapy has been shown to be effective and therefore reserved as a palliative approach[16,17]. In the present study, surgical therapy was determined to be the optimal treatment approach for

several reasons. The various treatment previously performed on the current patient did not result in an evident recession of the mass. Without surgical intervention, a firm mass and unbearable headache would remain.

CONCLUSION

The present study reports successful surgical treatment of a patient with a rare primary extracranial meningioma in the maxillary sinus. The present study demonstrated that imaging studies can aid in the diagnosis and biopsy and is useful to specify diagnosis. Surgical treatment is a viable option for the successful management of extracranial meningiomas in the maxillary sinus, and complete postoperative care often requires a multidisciplinary approach.

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