Sultan Qaboos University Med J, November 2024, Vol. 24, Iss. 4, pp. 575–577, Epub. 27 Nov 24
Submitted 21 Sep 23
Revision Req. 13 Nov 23; Revision Recd. 25 Nov 23
Accepted 19 Dec 23
https://doi.org/10.18295/squmj.12.2023.096

CASE REPORT

Palato-Antral Involvement of a Primary Extracranial Sinonasal Meningioma

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ABSTRACT: Primary extracranial sinonasal meningiomas are one of the rarest tumours involving the head and neck region. Very few reports exist with additional involvement of the palato-antral region. A 45-year-old male presented with frequent episodes of epistaxis, complaints of nasal blockage and nasal regurgitation for 8 months in 2021 at our tertiary care government hospital in East Godavari district, India. The patient was diagnosed with a primary extracranial sinonasal meningioma with palatal involvement and surgical excision via intra-oral and endoscopic approaches were performed. Postoperative healing was uneventful with no recurrence noted over 2 years. A palatal obturator was used for rehabilitation. The diagnosis of this pathology requires additional immunohistochemistry testing for confirmation and treatment entails complete surgical excision which assures no recurrence or delayed presentation of residual disease in follow-up.

Keywords: Meningioma; Paranasal; Maxilla; Palate; Case Report; India.

Meningiomas are known to be common slow-growing benign tumours of the central nervous system. Presentation of primary extracranial meningioma in the maxillo mandibular region is very rare with an incidence of 0.9–2%.[1–3] These are sporadic with suspected aetiology arising from the meningiocytes of the central nervous system via cranial nerves or as embryonic arachnoid cell rests.[4] Patients report complaints of nasal blockage, stuffiness, bleeding, painless mass, facial swelling, etc; this emphasises the importance of histological confirmation of the diagnosis to distinguish from other sinonasal or maxillary tumours and proceed with management. The prognosis of this benign tumour is favourable with disease recurring only if there is residual mass after definitive treatment.[5,6]

Case report

A 45-year-old male patient presented in 2021 to our tertiary care government hospital in East Godavari district, India, with complaints of nasal blockage, frequent episodes of epistaxis and nasal regurgitation for 8 months. The patient had no prior history of any allergies, sinusitis, rhinitis, trauma or headache. There was no history of any deleterious habit. Clinical examination revealed a soft swelling over the right cheek region and a palatal fistula with surrounding friable growth. Endoscopic evaluation showed a reddish mass in the right nasal cavity [Figure 1]. After routine blood investigations, biopsy and imaging were done to ascertain the nature of the mass. Contrast-enhanced computed tomography of the head and neck region was suggestive of enhancing soft tissue attenuating mass of 33 × 34 mm, involving the right naso-antral region with palatal erosion. No cranial pathology was noted [Figure 2A and B]. Biopsy from the tissue showed islands of spindle cells and fascicles of round cells with round nuclei showing a stippled chromatin pattern [Figure 2C]. Hyaline stroma with areas of necrosis was also seen in a few sections suggestive of grade II atypical meningioma. Bony trabeculae were noted to be invaded by these cellular areas. Immunohistochemistry was positive for vimentin, epithelial membrane antigen (EMA) and S-100 confirming the diagnosis of atypical sinonasal meningioma.

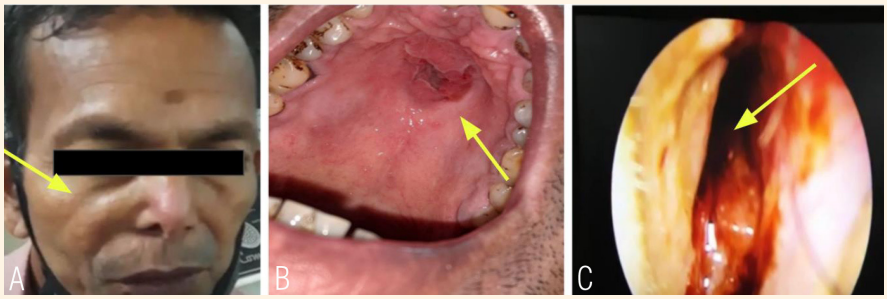

Figure 1: Photograph of a 45-year-old male patient with arrows showing (**A**) mild cheek swelling, (**B**) growth in palate with fistula and (**C**) an endoscopic view of mass in the right nasal cavity.

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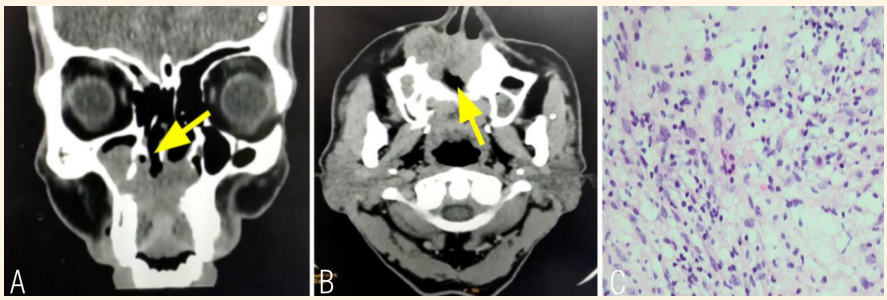

Figure 2: Image in the (**A**) coronal view - arrow depicting sino-nasal involvement (**B**) axial view- arrow depicting palatal bone erosion. Haematoxylin & eosin stain at ×400 magnification showing (**C**) islands of spindle cells, round cells with prominent nucleus and chromatin pattern seen.

Under general anaesthesia, a combination of intra-oral and endoscopic techniques was used for the excision. The vestibular approach exposed the tumour which was seen to be involving the anterior, antero-lateral wall, the floor of the maxillary sinus, the nasal floor and the palate. A complete excision of the tumour was done [Figure 3]. A titanium mesh was placed over the defect to provide support followed by closure. A surgical obturator was used for initial rehabilitation and was planned for a definitive prosthesis after healing. The patient was followed-up at regular intervals and was disease-free for 2 years [Figure 4].

Patient consent was obtained for publication purposes.

Discussion

Extracranial meningiomas are known to affect sites such as paranasal sinuses, orbit, scalp, ear and temporal bone. Primary extracranial sinonasal meningiomas are rare (i.e. without evidence of any intra-cranial pathology). Their origin is postulated to be as a direct extension of an intra-cranial meningioma via resorption of bone or arachnoid cell rests or as metastasis.[7] Literature shows a female predilection due to the presence of oestrogen, progesterone and androgen receptors on these tumours.[8] In the current case, the patient was a middle-aged male. Common presenting symptoms are nasal stuffiness, epistaxis, sinusitis, anosmia, headache, proptosis, periorbital oedema, etc; this is similar to any nasal benign or malignant mass. A differential diagnosis in cases with a sinonasal mass should include extracranial meningioma, which can be confirmed by a biopsy. The World Health Organization classifies this on a histological basis as grade I benign (consisting of the following types - fibroblastic, transitional, psammomatous and angiomatous; which account for 90% of meningiomas); grade II (consisting of atypical meningiomas which show hypercellularity, mitosis and necrosis, with occasional brain invasion); or grade III (malignant meningiomas which resemble melanomas, carcinomas or high-grade sarcomas with definitive cerebral invasion and rapid recurrence).[9] Strong reactivity for anti-EMA and vimentin are consistent with the diagnosis of meningioma, thus highlighting the importance of immunohistochemistry testing.[10] The current patient was diagnosed with atypical meningioma.

To determine the extent of the lesion, magnetic resonance imaging or contrast-enhanced computed tomography is generally undertaken. The current patient showed enhancement in the naso-antral region, with additional erosion of the palate and the maxillary wall. There was no breach into the orbital floor or any other cranial pathology. Involvement of maxilla-mandibular complex as a separate or an extension of sinonasal meningiomas has not been reported often.[2,4,11–14]

The definitive treatment for extracranial meningiomas is complete surgical excision with acceptable cure rates of 80–85% over 5 years.[14] Any recurrence is rare and is attributed to incomplete removal during the primary surgery. Most sinonasal meningiomas are excised by an endoscopic approach. However, due to the antrum and palate being additionally

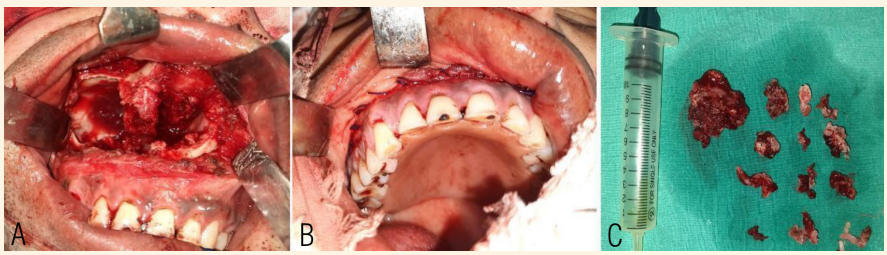

Figure 3: Photographs showing the (**A**) excision of mass from the antrum, nasal floor and cavity, (**B**) closure of incision and obturator in place and the (**C**) excised specimen.

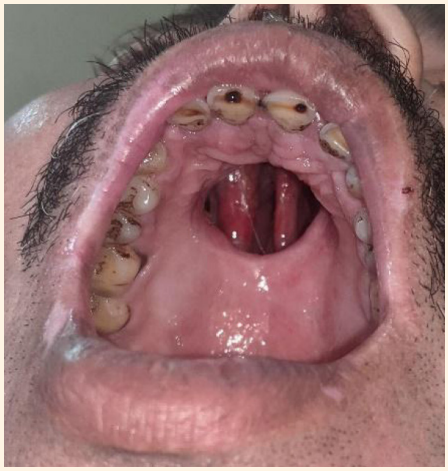

Figure 4: Follow-up photograph showing no residual disease.

involved in the current case, an intra-oral approach was used to remove the bulk of the tumour via the vestibular approach exposing the antral walls, pyriform rims, nasal floor and septum after initial endoscopic evaluation and excision. Closure was done after placing a titanium mesh for soft tissue support. Considering the patient's personal and economic preference, a surgical obturator was placed initially and later a definitive obturator was given, thus maintaining oral function and preventing nasal regurgitation.

A regular follow-up is advised especially in grade II meningiomas, with additional attention to any cerebral symptoms and subsequent associated imaging.[15] Healing was satisfactory in the current patient and he was disease-free during the follow-up period of 2 years. The patient was able to function well without nasal regurgitation during the consumption of food and had no nasality in his voice due to the palatal seal of the obturator. The 5-year disease-free survival rate ranges from 66.9–82.1%, and the 10-year disease-free survival rate ranges from 54.6–78.6%. The recurrence rate after complete resection varies from 7–84% according to follow-up.[16]

Conclusion

Primary extracranial meningiomas involving sinonasal, maxillary and mandibular sites are rare and require thorough diagnosis to identify the grade of tumour. Knowledge of this pathology is necessary for all maxillofacial and otolaryngologists as its presenting features are not uncommon. Adequate surgical excision by endoscopic or intra-oral or a combination of both gives satisfactory results and has good prognosis.

AUTHORS' CONTRIBUTION

VD managed the case, collected the data and drafted the manuscript. KPS and VD critically revised and edited the manuscript. Both authors approved the final version.

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