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Case Report

An atypical presentation of recurrent temporal lobe meningioma with external auditory canal mass

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Abstract

Extracranial spread of recurrent meningiomas involving the middle ear is rare. We present the case of a 59-year-old woman with headache and swelling of scalp over the right temporal region. MRI revealed a lesion in the right temporal lobe suggestive of meningioma. She underwent complete surgical excision of the lesion followed by post-operative radiotherapy. After 1 year, she presented with right-sided otalgia and a middle-ear mass extruding into the external auditory canal. She was re-operated and histopathology was anaplastic meningioma. We are discussing this unusual pattern of recurrence in our patient with a review of literature.

Key words: External auditory canal, recurrent meningioma

Introduction

Meningiomas account for 15% of all primary intracranial neoplasms. Recurrence of intracranial meningiomas after radiotherapy is uncommon and occurs usually in the primary site. Recurrent meningiomas involving the external auditory canal have been rarely reported. A 59-year-old woman presented with headache and swelling of scalp over the right temporal region. MRI revealed a lesion in the right temporal lobe suggestive of meningioma. She underwent complete surgical excision of the lesion followed by post-operative radiotherapy. She presented one year later with right sided deafness and a protruding auditory canal mass. Imaging revealed recurrence of meningioma.

Case Report

A 59-year-old female presented with gradually increasing swelling over the right temporal region of scalp of 1-year duration. She had no neurodeficit or any other symptoms except swelling over the scalp. CT scan revealed a well-defined mass measuring 4 × 4 × 4 cm in the right temporal region extending laterally and eroding squamous part of right temporal bone region and infiltrating right temporalis muscle. She underwent right temporal craniotomy with supratentorial approach and partial excision of tumor was done. Intra-operative findings were suggestive of a right temporal lobe mass with extension into adjacent right temporal bone and muscle. Floor of the middle cranial fossa was intact after surgery, thus preventing further spread of the disease to middle cranial fossa. The residual mass was in the deep right temporal lobe region. Though the epicenter of the lesion was suspected to be in the right temporal lobe, temporal lobe meningioma with brain involvement could not be ruled out. Histopathology showed features of anaplastic meningioma. She was diagnosed to be a case of anaplastic meningioma of right temporal lobe with extension to adjacent temporal bone and with post-operative minimal residual disease. She received post-operative three-dimensional conformal radiotherapy to partial brain with 6 MV linear accelerator with four non-coplanar fields to a dose of 60 Gy in 30 fractions in 6 weeks.

After 1 year, she presented with complaint of giddiness of 15 days duration and feeling of decreased hearing on the right side. On examination, she had excellent performance status with no neurodeficit, but had

right-sided conductive deafness with polyp-like mass protruding from right external ear [Figure 1]. Mass was cauliflower-like, brownish in color and non-tender with minimal discharge. CT scan showed a soft tissue mass in post-lateral part of right middle cranial fossa with extension into adjacent external ear and bulging into middle ear. MRI scan showed an enhancing mass in right temporal region and involving right squamous part of temporal bone, petrous bone and mastoid bone [Figure 2]. The mass was extending into external auditory canal (EAC) and around mandibular condyle. Complete surgical re-excision was done through both intracranial and extra-auricular approach. Histopathology revealed anaplastic meningioma. The patient had an uneventful post-operative recovery.

Discussion

Meningiomas account for 13-18% of all primary intracranial neoplasms. About 20% of intracranial meningiomas eventually develop an extracranial extension, most commonly to the orbit.[1] Following invasion of the temporal bone, the most common extension route is through the jugular and lacerate foraminae into the nasopharyngeal, retromaxillary, retromandibular and cervical spaces.[1] Meningiomas are known to be easily implanted and have a tendency to grow at the surgical scars where meningioma cells are deposited.[2] However, meningiomas involving the external auditory ear are very uncommon.[1] Meningiomas of the external auditory ear may be primary (very rare) or secondary due to extension of an intracranial meningioma.[1] Among the intracranial meningiomas, cerebello-pontine angle meningiomas have a tendency to involve the external auditory canal in the primary setting.[1] Most of the data regarding external ear meningioma were obtained from the pre-sophisticated imaging era, when there was little information regarding any silent intracranial disease. Retrospectively, it seems that there may be intracranial disease in few of these 'primary external ear' meningiomas, which was not detected by routine imaging.[3] Thus, primary middle ear meningioma is extremely rare and is to be suspected only after exclusion of any intracranial meningioma with extension. Despite the erosive pattern of growth and invasion to adjacent structures, external ear extension of intracranial meningiomas is not necessarily considered malignant and prognosis is considerably favorable.[2] The known female preponderance of intracranial meningiomas is also seen in external auditory canal meningioma. There is also an association with obesity in such cases.

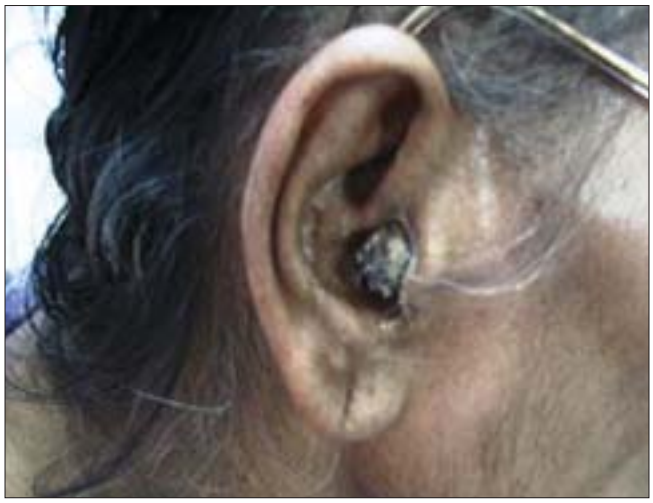

Figure 1: Clinical presentation of a recurrent temporal lobe meningioma as an external ear polyp

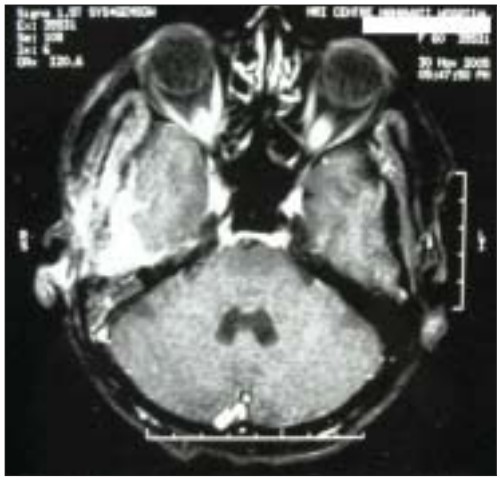

Figure 2: MRI scan showing a mass lesion in the right temporal lobe region extending into the auditory canal

Recurrence of intracranial meningiomas occurs usually in the primary site and less commonly at the surgical scar. Recurrent meningiomas involving the external auditory ear have been hitherto rarely reported.[2] Intracranial meningiomas with sinister histopathological features, like high mitotic index, nuclear pleomorphism (anaplastic meningioma) have higher probability of recurrence.[3] Younger patients with meningioma have a poorer outcome and higher local recurrence.[4]

Based on the previous reported series, surgery of a meningioma with EAC extension at presentation is the preferred treatment and is possible in most situations.[1] This rationale seems to hold true for recurrences too as was the case in our patient, even though the patient had received prior radiotherapy. Since the patient was irradiated before, we did not subject her to further radiation. In radiation-naïve patients, however, post-operative radiotherapy after recurrence in a meningioma can improve control rates.[5]

The differential of a lesion in the middle/external

ear needs to be ruled out in all instances of *de novo* and recurrent presentations since the prognosis of meningioma can be vastly different as compared to these lesions. Extension of meningiomas into the ear canal from an intracranial or skull-based location is well recognized. The importance of recognizing the lesion in this location rests in not confusing it with lesions that are more commonly encountered in this area, such as schwannomas, paragangliomas, squamous cell carcinomas, adnexal tumors, middle ear adenomas, ceruminous gland tumors, primary and metastatic carcinomas, and malignant melanomas. All these lesions have a more sinister course than a meningioma.[5,6] In a few cases, middle ear adenomas display endocrine differentiation and produce mucicarmin-positive material in the lumina. In addition, ceruminous gland-derived tumors in EAC (ceruminomas, adenoid cystic carcinomas and well-differentiated adenocarcinomas) may be considered a diagnostic alternative as well, but most have a typical histology and show myoepithelial differentiation in the basal cells. Mucoepidermoid carcinomas vary in their histologic appearance and may be included in the differential diagnosis. Some metastatic carcinomas may show hyaline material in the lumina, but atypia and mitoses appear in the context of a true epithelial tumor.[5,6]

In our case, the patient had temporal lobe anaplastic meningioma treated initially with complete surgical excision and post-operative radiotherapy with optimal doses. Despite this, she had recurrence at short interval, which could suggest an aggressive biology of the disease. The recurrence had occurred in an unusual site, the external ear canal. Recurrence could have been due to direct extension through the defect in the floor of the middle cranial fossa. This defect could be due to destruction of the middle fossa floor by the disease or iatrogenic after previous surgery.

To summarize, the management of recurrent meningiomas involving the EAC should initially involve surgical excision. Patients who have not received radiotherapy earlier can subsequently receive post-operative radiotherapy. In the case of the previously irradiated patient, the patient should be kept on follow-up only. The optimal management and the overall prognosis of such recurrences, in view of their rarity, are unknown.

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