CASE REPORT

The Meningioma that Got Cheeky

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Abstract Extracranial meningiomas are rare with a reported incidence of 1–2 %. Diagnosis is a challenge due to the unusual site of occurrence. The treatment of choice is surgical excision. A case that presented with primary lesion in the cheek with no detected intracranial extension is being reported.

Keywords Primary · Extracranial · Meningioma

Introduction

Meningiomas are tumors arising from the arachnoid villous structures of the brain or spinal cord. The incidence of extra-cranial extension of intra-cranial lesions is found to be 20 % with extension into the orbit, paranasal sinus, nasal cavity, parapharyngeal region, parotid region and outer table of scalp [1]. Even more rare are meningiomas that arise primarily in an extra-cranial location with 1–2 % incidence [1]. Such lesions are termed as ectopic, extra-calvarial or heterotrophic.

Only nine other cases have been reported in the jaw bones, to the best of our knowledge, of which six are mentioned by Taylor

[2] and three others by Yasutaka [3], Lell [4] and Elisabeth [5]. Apart from these cases Jin Woo Waeng et al. [6] have reported a meningioma presenting as a cheek mass and claiming a singularity in the english literature. Such a case of cheek swelling with no detected intracranial extension is presented.

Case Report

Sixty year female with a painless swelling of right cheek, of 30 months duration, which apparently developed following a tooth extraction. The swelling was approximately six by four centimetres extra orally extending from the lower border of mandible to temporal region superioinferiorly, corner of mouth to four centimetres behind it anterioposteriorly. Intraorally the swelling was four by three centimetres extending from third molar region to second premolar and depth of upper vestibule to three centimetres below it, irregular in shape. On palpation it was soft in the cheek but firm intraorally and in the temporal region (Figs. 1, 2). No tenderness, fluctuation or compressibility was elicited with no change in colour of overlying skin or mucosa. Pressure symptoms noted on the posterior aspect of maxilla with resultant tooth mobility. Skin and mucosa overlying the mass were not fixed. CT revealed a mass of heterogeneous density in the right cheek extending into retro-maxillary region and superiorly to the infra-temporal region with a medial extension along the inner aspect of the mandibular ramus. There was no delineation between the lesion and the masseter nor any bony erosion or invasion, though marked pressure effects were noted on the maxilla. Right maxillary sinus showed radiopaque changes regarding which ENT consultation was taken and opined as antral-polyp (Fig. 3). Biopsy of the lesion had been done twice elsewhere, slides were reviewed and the result was inconclusive ranging from lipoma, schwannoma to angiomeningioma.

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Fig. 1 Extraoral photograph of the lesion

Fig. 2 Intraoral photograph of the lesion

Surgical resection was carried out considering benign nature of the lesion based on the clinical history and radiographic findings via a combined lateral mandibulotomy approach and preauricular incision with temporal extension (Fig. 4). The histopathology was consistent with meningioma (Fig. 5). One year post resection the patient is doing well with no evidence of recurrence so far.

Discussion

Primary extra-cranial meningioma such as the one described above is very rare with only a few cases reported and only one such case presenting as cheek mass has been reported. The origin of such extra cranial primary meningiomas is controversial with various possible theories which include proliferation of the perineural cells of the nerves in the region; since perineurium is believed to be the direct extension of the arachnoid membrane [7]. Another theory proposing proliferation of ectopic arachnoid tissues along the course of cranial nerves has been put forward [8, 9].

The diagnosis of ectopoic meningiomas remains a challenge, the fact that such lesions are rare in the region and clinical and radiographic features of these tumors are non specific. Histopathologic confirmation also becomes a difficulty, due to the uncommon site, and immunohistochemistry plays an important role in the confirmation. CT scan and MRI are very important in determining the extent, bony involvement or breach, vascularization.

The extra-cranial meningiomas reported have a marked female predominance with nine of the twelve cases being females and the mean age was 49 years. Out of the reported cases in jaws and adjacent tissues most cases were in the age group of fourth to sixth decades with one case in a 10 year old male and one in a 26 year old female [3]. The extra-cranial meningiomas have been observed to be slow growing, benign asymptomatic masses. Some authors consider nerve injury during tooth extraction [3] as a possible stimulation. Other suggested stimuli may be trauma from RTA as reported by Maeng et al. [6].

The treatment of choice is surgical excision [2] and has good prognosis after complete resection with a median overall survival rate of 28.4 years. Lesion involving the jaws may require reconstruction following resection. Another important treatment modality is radiotherapy, which is been found to play an important role in the intracranial meningiomas in which complete resection was not possible [10]. Extra-cranial lesions can be managed with surgical clearance alone; radiotherapy is indicated only when there is an inoperable intracranial extension. Keeping in mind the limited number of cases reported so far regular long term follow up is advisable.

Conclusion

The diagnosis of extracranial meningioma based on clinical and radiographic findings is quiet obscure and confirmation requires histopathologic examination particularly immunohistochemistry. Surgical resection gives a good prognosis.

Fig. 3 CT windows showing lesion in infratemporal region. Note the pressure effects on the maxilla with no breach in walls

Fig. 4 Extirpated lesion

Fig. 5 Photomicrograph showing plump ovoid spindle cells with vesicular nuclei proliferating in the form of sheets and whorls in collagenous connective tissue stroma

Conflict of interest No conflict of interest declared.

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