Schwannoma in Infratemporal Fossa in a Young Patient: A Case Report

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Introduction

Neurogenic tumors of head and neck are uncommon in children [1]. Schwannomas are benign, encapsulated peripheral nerve tumors arising from the Schwann cell [2]. It was first reported by Verocay in 1910. Approximately 37–45 % of schwannomas occur in the head and neck region, and the lesions commonly arise from the roots of cervical and cranial nerves in the parapharyngeal space, with the majority originating from the vagus nerve [3, 4]. Majority of schwannoma are seen parapharyngeal space. Their occurrence in infratemporal space is very rare [5, 6]. Trigeminal schwannomas account for about 0.2 % of all intracranial tumors and for about 2–3 % of all intracranial schwannomas [4]. They may arise in any segment of the nerve, but the majority develops at the gasserian ganglion. This article reports a case of a 10 year old boy diagnosed with schwannoma in infratemporal fossa and treated by complete resection of the lesion by combination of transzygomatic and intraoral approach.

Case Report

A 10 year boy reported in the out patient department of Government College of Dentistry, Indore with a chief complaint of progressively increasing swelling below left eye (Fig. 1) for the past 4 months with associated pain for the past 3 months. His parents noticed widening of palpebral fissure of left eye compared to the right eye. There was

no history of trauma, blurring of vision, nasal blockage or epistaxis. Left eye was enlarged for the past 3 months. No significant family or medical history was reported. Clinical examination showed ear, nose and throat to be within normal limits. There was proptosis of left eye medially and superiorly. Eyeball movements were normal vision was 6/6 in both eyes.

The swelling was diffuse and was approximately 4×4 cm with no signs of inflammation. The swelling was extending from left infratemporal region to mid of cheek and encroaching the left vestibule intraorally. Paranasal sinus radiograph also reveals haziness in left maxillary sinus. Computerised tomography (CT) scan showed large expansile, heterogeneously enhancing mass in infratemporal fossa (ITF) measuring $5.4 \times 4.4 \times 4.5$ cm. Scalloping of inferior orbital wall was appreciated, causing mild proptosis (Figs. 2, 3).

Laterally, the swelling was limited by zygomatic arch and extending into posterior half of nasal cavity. Scalloping of maxillary sinus wall is also seen in axial view of CT scan (Fig. 4).

Biopsy was taken intraorally from buccal vestibular area. Biopsy report showed sections of spindle shaped with neural differentiation tumour cells arranged in hypercellular Antoni A and hypocellular Antoni B pattern. Verocay body also noted (Fig. 5).

Histopathological Diagnosis: Schwannoma

Surgical plan included complete excision of the tumor. Because of large anteroposterior extensions of the tumor in infratemporal fossa, transzygomatic approach was considered (Fig. 6). A hemicoronal incision extending up to the lobule of ear was made through skin, subcutaneous tissue and galea. Superficial layer of temporal fascia was incised

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Fig. 1 Clinical presentation of the patient showing facial asymmetry

Fig. 2 CT showing a well encapsulated mass with heterogenous enhancement in left infratemporal fossa

Fig. 3 CT revealing scalloping of left infra orbital margins

Fig. 4 Axial view CT showing scalloping of maxillary sinus wall

in temporal region. A 45° angle incision was made at the root of zygomatic arch, incising the superficial layer of temporalis fascia. After incising the periosteum, zygomatic arch was exposed. Zygomatic arch was osteomized, hinged inferiorly and reflected on masseter. Coronoid process was

Fig. 5 Microscopically, hypercellular areas of spindle-shaped cells with nuclear palisading pattern (hematoxylin eosin stain; original magnification $\times 40$)

Fig. 7 Clinical photograph of excised tumour mass

Fig. 6 Zygomatic arch osteotomy performed and tumor mass exposed

sectioned and temporalis was reflected superiorly exposing the tumor mass. Intraoral maxillary vestibular incision was made to completely identify the inferior extent of the tumor. Complete excision of the tumor was performed (Fig. 7). After removal of tumor, zygomatic arch was brought back

Fig. 8 zygomatic arch was repositioned in anatomic position and plating done

to its anatomic position and fixation was performed with titanium plates (Fig. 8). The post-operative period was uneventful. At 1 year regular follow up, the patient is presenting with good facial symmetry and marked decrease in proptosis in left eye (Fig. 9) There is no evidence of recurrence of tumor, although a CT scan, which is an ideal method to rule out recurrence was not carried out.

Fig. 9 1 year post operative clinical photograph revealing marked decrease in facial asymmetry

Discussion

Head and neck region accounts for approximately 37–45 % of all solitary schwannomas. Schwannomas are most common in 2nd–3rd decade of life and there is no sex predilection [8]. Clinically they are associated with pain at the site of concerned nerve. Features like proptosis; swelling is predominantly determined by the extension of the neoplasm. Bone erosion is usually secondary to pressure necrosis than by invasion. In these regions, schwannomas are most frequently found in the parapharyngeal area in association with lower cranial nerves or the sympathetic chain [11]. The infratemporal fossa is one of the least common anatomical sites for schwannomas [9, 10]. The infratemporal fossa is a quadrangular space bounded anteriorly by the maxillary sinus, medially by the pterygoid plate, laterally by the temporalis muscle and ramus of the mandible, and superiorly by the greater sphenoid wing in the floor of the middle cranial fossa. It is important to note that the upper gingivo-buccal sulcus forms the floor anteriorly and masses in the infratemporal fossa may present first in this region [12]. The infratemporal fossa contains the medial and lateral pterygoid muscles, the mandibular nerve and its branches, the first and second parts of the maxillary artery and the accompanying

veins, the otic ganglion, the vidian nerve and the sphenomandibular ligament [11]. Sometimes it may also contain salivary glands which may develop tumors which present in this region [13]. Medially, the infratemporal fossa communicates with the pterygo-palatine fossa through the pterygomaxillary fissure. Medially, the superior constrictor muscle separates it from the nasopharynx. Posteriorly, the infratemporal fossa is continuous with the parapharyngeal space. Anteriorly, this space communicates with orbit through the inferior orbital fissure. This space also communicates with the temporal fossa through a wide opening under the zygomatic arch. Surgical removal of symptomatic schwannomas of infratemporal fossa is done by various surgical approaches depending on the location of the tumor. The tumor mass can be approached intraorally by either isolated maxillary vestibular or Caldwell Luc approach. This approach has a limitation of insufficient exposure. Tumor extension into the orbit or the middle cranial fossa is difficult to treat with this exposure. Anterolateral route includes frontotemporal orbitozygomatic and transcranial subtemporal frontotemporal approach. They give a very good exposition of the infratemporal fossa, but by paying the price of craniotomies and high invasiveness. Anterior route is transmaxillary approach using Weber Fergusson or Caldwell Luc approach. This approach has a disadvantage of scar formation at the incision site and maxillectomy. Minimally invasive endoscopic route includes transnasal and transmaxillary approach [7]. Lateral route is combined transzygomatic and maxillary vestibular approach. Owing to the large size of tumor (approximately $5.4 \times 4 \times 4.5$ cm) a combined tranzygomatic and intraoral approach was considered necessary to remove the tumor entirely. This transzygomatic approach is similar to one described by Kumar [14]. After coronoidectomy, temporalis muscle was retracted superiorly to gain sufficient access to the tumor mass. This approach was more aesthetically acceptable to the patient and is not associated with any post operative morbidity. At long term follow up, patient is asymptomatic with good facial symmetry.

In conclusion, schwannomas limited to ITF can be effectively excised by combined transzygomatic and intraoral approach. Not only schwannomas, this approach can be used equally effectively in other lesions in ITF, but further accumulated data or patients are needed to support the effectiveness of this approach.

Conflict of interest None.

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