Pleomorphic Adenoma in the Infra-temporal Space: The First Case Report

K. Jeyanthi · Ramalingam Karthikeyan · Herald J. Sherlin · N. Anuja ·
Pratibha Ramani · Premkumar Priya · T. Chandrasekar · Naushad Khan ·
Vinod Narayanan

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Abstract The infratemporal fossa is a deep retromaxillary space corresponding to the inferior aspect of the middle cranial fossa. Tumours in the infratemporal fossa are very rare; therefore experience in their diagnosis and therapeutic management is very limited. Tumors that are reported commonly in the infratemporal region are nasopharyngeal carcinomas, juvenile angiofibromas and adenoid cystic carcinomas. To the best of our knowledge, this is the first report in the English language literature of a pleomorphic adenoma presenting in the infratemporal space and probably arising from an ectopic minor salivary gland, along with a brief description of the surgical technique.

Keywords Pleomorphic adenoma · Infratemporal fossa

Introduction

The infratemporal fossa is a deep retromaxillary space corresponding to the inferior aspect of the middle cranial fossa [1]. The infratemporal space is an anatomic region located behind the maxillary tuberosity, bounded anteriorly

by the maxillary tuberosity, posteriorly by the lateral pterygoid muscle, the condyle and temporal muscle, laterally by the tendon of the temporal muscle and the coronoid process, and medially by the lateral pterygoid plate [2]. Tumors in the infratemporal fossa are very rare; therefore experience in their diagnosis and therapeutic management is very limited.

Pleomorphic adenomas account for 40–70% of minor salivary gland tumours. They may arise in unusual locations along the cheek, along the Stenson's duct and in accessory parotid tissue, that is separated from the body of the gland [3], in the parapharyngeal space arising either *denovo* or from the deep lobe of the parotid gland and extending through the stylomandibular tunnel into the parapharyngeal space [4]. It may also develop in salivary gland inclusions within the lymph nodes in the neck [3].

To the best of our knowledge, this is the first report in the English language literature of a pleomorphic adenoma presenting in the infratemporal space and probably arising from an ectopic minor salivary gland [5]. A brief description of the surgical technique is presented.

Case Report

A 45-year-old Asian Indian female patient presented with fullness on the right side of the face, first noted approximately 6 months prior to presentation. She also experienced occasional pain in the region for the past 2 months. The patient's past medical history was non-contributory.

Extra-oral examination revealed a mild facial asymmetry of the right cheek region. The skin over the region was not fixed to any underlying structures and was freely movable in all planes. There was no evidence of regional

K. Jeyanthi · R. Karthikeyan · H. J. Sherlin · N. Anuja ·
P. Ramani (✉) · P. Priya · T. Chandrasekar
Department of Oral and Maxillofacial Pathology,
College of Dental Surgery, Saveetha University,
162, P.H. Road, Velappanchavadi,
Chennai 600077, Tamilnadu, India
e-mail: drrkn@rediffmail.com

N. Khan · V. Narayanan
Department of Oral and Maxillofacial Surgery,
College of Dental Surgery, Saveetha University,
162, P.H. Road, Velappanchavadi,
Chennai 600077, Tamilnadu, India

lymphadenopathy. Her mandibular opening was within the normal range (25 mm).

Intra-oral examination revealed a firm, nodular swelling obliterating the right buccal vestibule, extending from the zygomatic buttress adjacent to tooth 16 region and the maxillary tuberosity (Figs. 1 and 2). On palpation, the swelling was tender. The overlying mucosa was free of any surface change, ulceration, purulent discharge or sinus tract. The tongue, floor of the mouth and hard and soft palate were all normal.

Serial direct coronal and axial sections of the paranasal sinuses were studied without administration of I.V. contrast. It revealed a moderate sized isodense lesion ($3.1 \times 2.2 \text{ cm}^2$) in the right retromaxillary space, with

Fig. 1 Extra-oral picture showing mild facial asymmetry on the right cheek region

Fig. 2 Intra-oral picture showing obliteration of the buccal vestibule

anterior bowing of the posterolateral wall of the adjacent right maxillary antrum. The lesion was closely related to the temporalis muscle and there was no destruction or erosion of adjacent mandibular or maxillary antral wall. Nasal septum was in the midline, nasal turbinates, superior, middle and inferior meati, maxillary ostium, bony walls of all the sinuses, nasopharynx, both orbits and its contents were normal. The radiologist has opined that it is a well-encapsulated solid tumor in the infra-temporal fossa, separate from the parotid gland indenting upon the posterolateral wall of the right maxillary antrum (Fig. 3).

Fig. 3 Well-encapsulated solid tumor in infra-temporal fossa, separate from the parotid gland indenting upon the posterolateral wall of the right maxillary antrum

Based on the clinical and radiologic findings, the provisional diagnosis given by the clinicians was fibroma or angiofibroma.

An incisional biopsy was taken from the right buccal vestibular region, near the upper first molar and sent to the Department of Oral Pathology, for evaluation. Histopathology revealed an encapsulated lesion comprised of duct like spaces with an eosinophilic coagulum, surrounded by spindle shaped and plasmacytoid cells in a dense, hyalinized connective tissue stroma. An area resembling chondroid tissue was also evident. The histopathological features were suggestive of pleomorphic adenoma. (Figs. 4 and 5).

Subsequently, surgery was planned with a zygomatic arch access osteotomy and coronoidotomy to excise the tumor mass, from the infratemporal fossa.

Fig. 4 Histopathological picture showing an encapsulated lesion comprising of duct like spaces with eosinophilic coagulum (10 $\times$, H&E stain)

Fig. 5 Histopathological picture showing an area resembling chondroid tissue (10 $\times$, H&E stain)

A Hemicoronal incision with pre-auricular extension was used to expose the zygomatic arch on the right side. The osteotomy cut was placed at the root of the zygomatic arch. A Weber-Fergusson incision was used to expose the intra-oral part of the tumor. Tumor mass along with the capsule was dissected up towards the zygomatic arch. Another osteotomy cut was placed on the zygomatico-maxillary suture. The osteotomized zygomatic arch was elevated with the flap. Coronoidotomy was done to elevate the temporalis muscle from the tumor mass (Figs. 6 and 7). Then, the total tumor mass was dissected out from the infratemporal fossa. The zygomatic arch was fixed with miniplates and the coronoid was fixed with transosseous wiring.

The excised tumor mass was sent for histopathological evaluation. On gross examination, the tumor mass measured 6 $\times$ 5 cm² in size, with a whitish, faintly lobulated and focally glistening cut surface. The histopathological findings of the excised specimen were consistent with the incisional biopsy report, confirming the diagnosis of pleomorphic adenoma.

The patient is on a regular follow up and is disease free till date.

Fig. 6 Picture showing the modified surgical approach performed with a zygomatic arch access osteotomy and coronoidotomy

Fig. 7 Picture showing the tumor dissected out from the infratemporal fossa

Discussion

Tumors of the infratemporal fossa are rare. Because of the anatomic location of the infratemporal space, these tumors remain silent for a long time. The frequent clinical presentation of the tumors in the infratemporal fossa has been reported in the superior gingivobuccal sulcus. Similarly, our patient also presented a swelling in the maxillary buccal vestibular region [1].

Extracranial tumors of the infratemporal fossa are considered to be of three types: namely, primary, contiguous and metastatic. Facial pain is frequently a presenting symptom of these tumors, many months before the diagnosis [5]. Tumors that are most common in the infratemporal region are nasopharyngeal carcinomas, juvenile angiofibromas and adenoid cystic carcinomas [6]. Pleomorphic adenoma (PA) arising in the infratemporal region has not been reported in the English literature to the best of our knowledge.

CT scan is an important diagnostic tool in tumors of the infratemporal fossa because it helps in determining the extent of disease, local spread and also helps to some extent in determining the type of tumor. Contrast enhancement is seen in vascular and neurogenic tumors. If a vascular tumor is suspected, an MRI angiography or conventional arteriography should be performed. If the possibility of a vascular lesion is ruled out, a histologic study could be performed [7]. The histopathology of our case proved to be pleomorphic adenoma.

Though there is an anatomic relationship between the deep lobe of the parotid gland and the infratemporal fossa, the probability of deep lobe tumors spreading to the infratemporal fossa seems to be rare. The possibility of a

superficial lobe tumor invading through the mandibular notch into this region was ruled out by CT scan images and surgical details. The extension of tumors from the deep lobe of parotid gland is distinguishable from tumor arising *de novo* in the infratemporal region by a fine lucent line representing the compressed layer of fibroadipose tissue between the tumor and deep lobe of parotid [8]. All these findings indicate that the tumor has arisen from an ectopic minor salivary gland within the infratemporal fossa.

The most common site of a PA of the minor salivary glands is the palate followed by lip, buccal mucosa, and floor of the mouth, tongue, tonsil, pharynx, retromolar area and nasal cavity [8, 9].

Pleomorphic adenoma in the parapharyngeal space can arise either *de novo* or may arise in the deep lobe of the parotid gland and extend through the stylomandibular tunnel into the parapharyngeal space. Tumors in the parapharyngeal space are rare and constitute less than 0.5% of head and neck neoplasms. Of these, PA is the commonest benign tumor (40%) [10].

The elective treatment of a pleomorphic adenoma is surgery. There are numerous surgical approaches, indicating the difficulty of access, accentuated by the communications with the neighboring regions. The main obstacles approaching the infratemporal fossa are the zygomatic arch, the parotid gland, the facial nerve and the ascending ramus of the mandible [11]. Thus for our case, a modified surgical approach was performed with a zygomatic arch access osteotomy and coronoidotomy to excise the tumor mass from the infratemporal fossa.

The prognosis of a pleomorphic adenoma is good. Because of its location in the infratemporal fossa, clinical monitoring is not sufficient during follow-up and periodic radiological evaluation is needed to detect any recurrence. The patient is remaining disease free after surgical excision and is on a regular follow-up.

Conclusion

In summary, we are reporting the first case of pleomorphic adenoma presenting in the infratemporal fossa.

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