Solitary fibrous tumor of the infratemporal fossa : A case report

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Abstract

First described as a pleural neoplasm, the solitary fibrous tumor (fibrous mesothelioma) has been reported in a number of extrapulmonary sites, including the Head-Neck region. In the Head-Neck region, it has been described in the sinonasal tract, epiglottis, parapharyngeal, retropharyngeal spaces, parotid and infratemporal fossa. We present the second case of solitary fibrous tumor of infratemporal fossa described in world literature. A complete excision was achieved by transmaxillary approach.

Keywords Solitary fibrous tumor • Fibrous mesothelioma • pleural neoplasm • Infratemporal fossa.

Introduction

Solitary fibrous tumor also called fibrous mesothelioma is referred to a distinctive neoplasm, which most frequently occurs in the pleura and was first described in 1931¹ and less commonly in relation to other serosal surfaces such as peritoneum. Recently these tumors have been documented in a variety of extrapleural site including mediastinum, lung, liver, sinonasal tract, epiglottis, parapharyngeal space.

The histogenesis of solitary fibrous tumor is controversial. Many recent immunohistochemical and ultrastructural studies argue strongly in favour of mesenchymal cell origin which is supported by the occurrence of solitary fibrous tumor in locations away from serosal surface^{2,3}. Rayappa⁴ et al reported the first case of solitary fibrous tumor of the infratemporal fossa (IFT) in 1996. Here we report the second case of solitary fibrous tumor arising in infratemporal fossa which was excised completely through transmaxillary approach.

Case History

A 35-year-old previously healthy male patient, a Doctor by profession, presented to us with history of pain and loosening of first and second maxillary molar tooth with fullness in cheek on the right side. This was associated with occasional trismus and fatigue of upper jaw especially after meals. Patient also gave history of pain and paresthesia over right side of the face of 3 month duration and extraction of maxillary molars outside before coming to us. Physical examination revealed slight fullness over the cheek and small ulcerated area over upper right alveolus.

There was no loss of sensation over the face. A contrast enhanced CT scan showed a smooth enhancing lesion in right pterygopalatine fossa and infratemporal fossa displacing the posterior wall of maxillary antrum anteriorly (Fig 1). Patient had undergone biopsy from the ulcerated area over the upper jaw outside and the biopsy was reviewed

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Fig. 1 Contrast enhanced computerized tomography axial section showing contrast enhancing homogenous mass in right infratemporal fossa, displacing the posterior wall of maxillary antrum anteriorly and infiltrating the pterygoid muscle. There is also soft tissue density in both maxillary sinuses.

Fig. 2 Per-operative photograph showing anastomosis of infraorbital nerve. The site of anastomosis shown by two black arrows.

Fig. 3 Photomicrograph of the solitary fibrous tumor showing spindle cell in a myxoid fibrous stroma (H & E; 10X).

at our institution but a definite histopathological diagnosis could not be made preoperatively. A possibility of benign spindle cell tumor was kept.

The tumor was excised utilizing a transantral approach. Weber Fergusson incision was made without lip split and subperiosteal flap elevated. The infra orbital nerve was identified and was divided in beveled fashion. Both the cut ends were tagged with a suture for further identification. The zygomatic bone was not removed to preserve the contour of the face. A wide anterior maxillary wall removal was initially accomplished. Posterior wall of maxillary sinus was found to be thinned out and ballooning into the maxillary sinus. There was firm, grayish white, multilobulated bleeding mass firmly attached to surrounding tissue present in right ITF dissected out by blunt dissection and removed in toto. The bleeding was controlled with bipolar diathermy. At end of the surgery the cut end of infraorbital nerve was approximated by 10 to 0 prolene suture (Fig. 2).

The post operative period was unremarkable. The histopathology of excised specimen confirmed the diagnosis of solitary fibrous tumor. Histopathologically the tumor showed a relatively well circumscribed tumor which was predominately capsulated. The tumor cells were mainly spindle in shape and arranged in short interlacing fascicles with numerous vascular channels (Fig. 3). Focal haemangiopericytoma like areas were also seen. The spindle cell population of the tumor demonstrated diffuse positivity with CD34 and Bcl2 immunostain. The patient is asymptomatic 6 months after surgery without any evidence of recurrence. The sensation over the right cheek improved over time and came normal 3 months after the surgery.

Discussion

Solitary fibrous tumors are rare neoplasms that most commonly occur as pleural or serosal tumors. Solitary fibrous tumors in pleura have a unique gross and microscopic appearance which has been well documented. The situation is different when it affects extrapleural sites. Ours is the second case to be reported in the ITF. It usually occurs in adults, age ranges from 30 to 64 years and both sexes are equally affected. Clinically, extrapleural solitary fibrous tumor may present with symptoms related to the site of origin of tumor or with systemic symptoms such as hypoglycemia, arthralgia, osteoarthopathy and clubbing of fingers^{5,6,7}. The systemic symptoms usually resolve on removal of the tumors⁷. Our patient did not have any systemic symptoms. Frequently, these tumors are detected incidentally on radiography or at surgery for some other reason. The histogenesis of solitary fibrous tumor is controversial. Klempere and Rabin¹ first hypothesized a mesenchymal origin but Stout and Murray⁷ subsequently proposed a mesothelial origin. However, the occurrence of solitary fibrous tumor in extraserosal site and the result of many recent immunohistochemical and ultrasrucural studies strongly suggest mesenchymal origin.

Macroscopically, these tumors appear well circumscribed and cut surface are generally pale and firm. Microscopically, the tumor is composed of varying number of spindle cell arranged in a collagenous background of variable vascularity. Occasionally the vascular spaces formed a branching network in a hemangiopericytoma like pattern. These tumors have two fundamental architectural features: solid spindle and diffuse sclerosis⁸ and both are found in same tumor in different proportion. The solid types are frequently confused with (1) vascular tumor like hemangiopericytoma, angiofibroma, (2) neurogenous tumors like schwannoma, (3) sarcoma like fibrosarcoma (4) various types of fibrous histiocytomas⁹. In most reports the tumor cells stain for vimentin and not for keratin, epithelial membrane antigen, desmin, carcinoembryonic antigen and S-100^{6,10}. The clinical behavior of the tumor is unpredictable. The aggressive clinical behavior is manifested by 13% to 23% of pleural solitary fibrous tumors^{3,11}.

This usually takes the form of local invasion and recurrence, although distant metastases have occasionally been documented³. Long term follow up is required as the recurrence can occur up to 30 years after surgery. The putative indicators of aggressive behavior, thought to be associated with large size, hypercellularity, increased mitosis, pleomorphism and hemorrhage, but these are unreliable^{3,11}. Tumors with a benign microscopic appearance have behaved aggressively and some with a malignant appearance behaved benignly⁶. Resectability is the most important prognostic factor. The management of solitary fibrous tumor is complete surgical excision.

Many surgical approaches have been described for removal of tumor from the infratemporal fossa. Rayappa⁴ et al described orbitozygomatic infratemporal craniotomy approach for excision of tumor. It required hemicranial incision, elevation of frontozygomaticotemporal bone flap and exposure of bifurcation of the common carotid artery. We feel transmaxillary approach to be ideal as it gives head on approach to the lesion.

The anaesthesia of the cheek injury to infraorbital nerve can be obviated by end to end anastomosis as was done in our case. Some authors have reported good result with radiation therapy or chemotherapy for incompletely excised tumor⁶. It is very difficult to come to any conclusion regarding any prognostic indicator because of rarity of this tumor in head neck region. In treatment planning and follow up we should assume that the behaviour of SFT in head neck may mirror the behaviour in other areas.

Key Message: Our case calls attention to a rare solitary fibrous tumors arising in ITF that must be considered in differential diagnosis of spindle cell lesion of ITF. Recognition of the occurrence of this tumor in the ITF is necessary to avoid erroneous diagnoses like hemangiopericytoma fibrosarcoma, which have many adverse consequences for patient.

Summary

We report the second case of solitary fibrous tumor of infratemporal fossa. The tumor was excised through transmaxillary approach as it gives head on view of the lesion. Our case also highlighted the fact that the anesthesia of cheek secondary to injury to infraorbital nerve can be prevented by end to end anastomosis of the nerve.

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