



Solitary Fibrous Tumor Occupying the Temporal and Infratemporal Fossa: A Rare Case Report

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Abstract

Solitary Fibrous Tumors (SFTs) are an uncommon mesenchymal neoplasm, mainly treated by surgical resection. A complete resection is important, but it may be difficult to secure sufficient margins with preserved functionality and esthetics in the head and neck region. We report a rare case of SFT occupying the temporal and infratemporal fossa. A 36-year-old woman presented with a tumor in the right temporal and infratemporal fossa. A core needle biopsy was performed; histopathological examination led to a diagnosis of SFT. We resected the tumor with a hemifacial dismasking flap and a zygomatic approach; reconstruction was performed using a free anterolateral thigh flap. The resected tumor revealed negative surgical margins on histopathology, with no recurrence at 1 year postoperatively. A facial dismasking flap is useful for approaching deep face areas, such as the infratemporal fossa or skull base. Moreover, the upper two-thirds of the face can be unfolded without any noticeable facial wounds, and the facial nerve can be preserved. Because the visual field could be secured only by temporary zygomatic arch transection and mandible coronoid process transection, we selected a zygomatic approach. We considered the facial dismasking flap and zygomatic approach as useful given the tumor localization and postoperative esthetic outcome in this case.

Keywords: Solitary fibrous tumor; Head and neck; Infratemporal fossa; Facial dismasking flap; Zygomatic approach

Introduction

Solitary Fibrous Tumors (SFTs), an uncommon mesenchymal neoplasm, comprise an intermediate group of tumors in the WHO classification that occur in all parts of the body, and were first described in the pleura by Klemperer and Rabin in 1931 [1]. SFTs commonly develop at 50 to 60 years of age, without a sex difference [2,3]. SFT incidence is greatest in the thoracic cavity (pleura, lung) at 68%, followed by the pelvis at 16%; occurrence in the head and neck is relatively rare, at 6% [2]. In the head and neck region, SFTs are most common in the buccal mucosa, followed by the nasal cavity, pharynx, tongue, and orbit; few case reports of SFTs in the temporal fossa exist [4]. Herein, we report a rare case of SFT occupying the temporal and infratemporal fossa resected using a hemifacial dismasking flap.

Case Presentation

A 36-year-old woman gradually experienced temporal swelling without pain for 3 years and was referred to our institute. At the first visit, an elastic hard mass was found in the right temporal region (Figure 1a). She had no apparent cranial nerve palsy. Contrast-enhanced Computed Tomography (CT) showed a well-defined and well-contrasted tumor (size, 45 mm × 24 mm × 51 mm) located from the temporal fossa to the pterygopalatine fossa. No obvious bone infiltration was observed in the cheekbones, skull base, or mandible (Figure 1b). Magnetic Resonance Imaging (MRI) revealed a tumor with good contrast on contrast-enhanced T1-weighted fat-suppression imaging (Figure 1c, 1d). Although the temporal and pterygoid areas were displaced and mastication muscles were thinned, the tumor did not infiltrate the surrounding tissues, including the parotid gland.

We performed a core needle biopsy before surgery. Histopathological examination showed round and spindle-shaped cells arranged irregularly (i.e. patternless pattern). Immunohistochemical staining revealed that the tumor cells expressed STAT6 and CD34 (Figure 2). The tumor cells also expressed CD99, bcl2, and S100, but not SMA and desmin. Thus, the tumor was diagnosed as an SFT, and surgery was performed approximately 8 weeks after the initial diagnosis.

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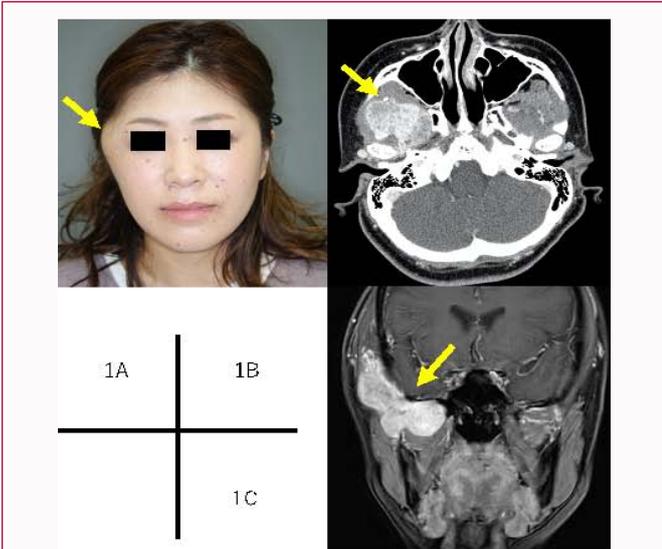


Figure 1: a) An elastic hard mass is observed on the right side of the temporal region; b) Contrast-enhanced computed tomography image (Axial); c) Post-contrast Magnetic Resonance Imaging (MRI) T1-weighted fat suppression image (Coronal). A well-defined tumor with a contrast effect is located from the temporal fossa to the pterygopalatine fossa. No obvious bone infiltration is observed in the cheekbone, mandible, or sphenoid bone. The surrounding masticatory muscles are thinned, but no obvious infiltration is observed.

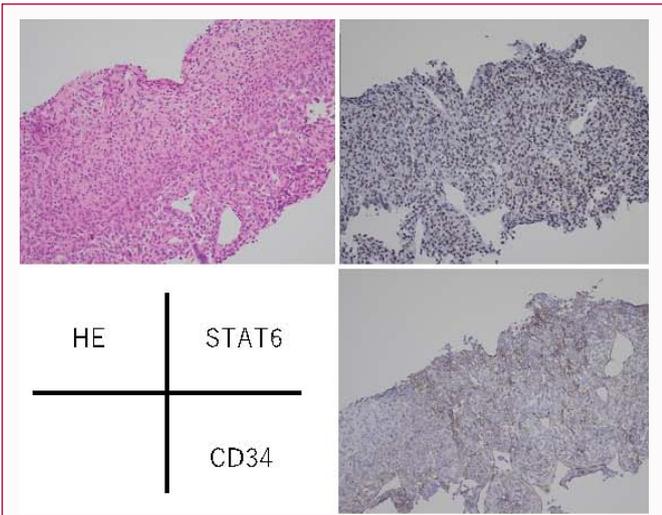


Figure 2: Histopathological findings (core needle biopsy). Hematoxylin and eosin staining revealed a patternless pattern, with round and spindle-shaped cells are irregularly arranged. Immunostaining revealed STAT6 and CD34 as positive.

For tumor resection, we used a hemifacial dismasking flap, with combined coronal skin and unilateral circumpalpebral incisions. The flap was raised under the periosteum to preserve the facial nerves and muscles. By adding a circumpalpebral incision, the flap could be detached from orbital structures (Figure 3a). The tumor was resected using a zygomatic approach. We temporarily transected the zygomatic arch, and the temporal and lateral pterygoid muscles were resected as surgical safety margins (Figure 3b). As the tumor had spread to the pterygopalatine fossa, the pterygopalatine artery, pterygopalatine ganglion, and mandibular nerve were sacrificed under the skull base (Figure 3c). Following tumor resection, we performed maxillofacial reconstruction using a free anterolateral femoral flap with superficial

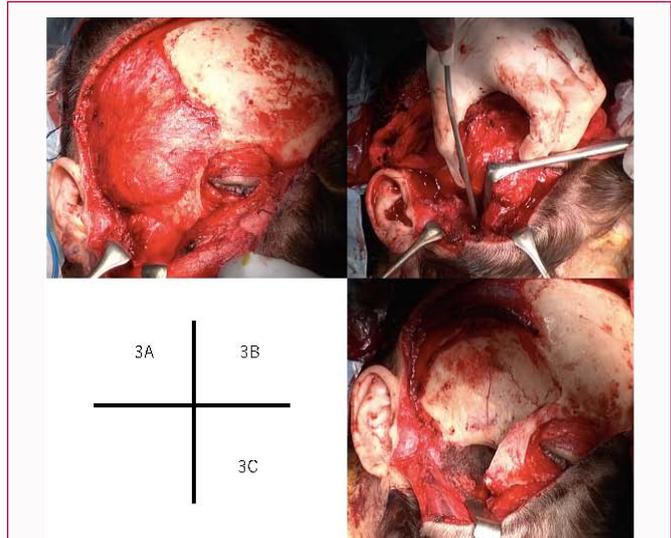


Figure 3: Surgical findings: a) With the hemifacial dismasking flap raised, the cheekbone, orbital contents, and temporal muscle can be confirmed, and tumor swelling (dotted line) is observed. The cheekbone was cut as shown in the figure (solid line). b) The zygomatic arch and coronoid process are removed, and the tumor is grasped and towed. The temporal muscle is transected, and the lateral pterygoid muscle is dissected. c) Post-excisional findings: with the zygomatic arch and coronoid process removed, and the tumor removed along with the temporal muscle, lateral pterygoid muscle, and skull periosteum, venous bleeding from the pterygopalatine fossa is observed, and cotton-like oxidized cellulose is placed to stop the bleeding.

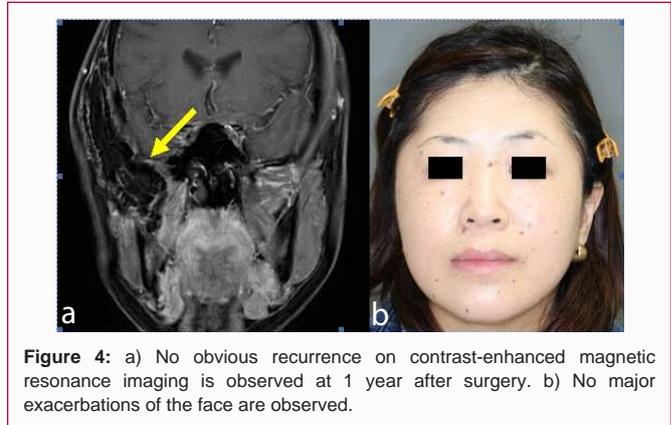


Figure 4: a) No obvious recurrence on contrast-enhanced magnetic resonance imaging is observed at 1 year after surgery. b) No major exacerbations of the face are observed.

temporal artery and vein anastomoses. The transected zygomatic arch was then returned and fixed. The surgery time was 6 h 20 min (bleeding, 1,014 ml).

Histopathological examination of the resected tumor revealed negative surgical margins. Immunohistochemical staining showed the same result as the preoperative biopsy, and the MIB-1 labeling index was <1%. The final pathological diagnosis was SFT.

Discussion

Her facial appearance showed no major problems postoperatively (Figure 4a) and she had a good course, with no recurrence on MRI at 1 year postoperatively (Figure 4b).

tumor shows hypo-intensity on T1-weighted images and various signal intensities on T2-weighted images. Infiltrative growth into the surrounding tissues is rare, but can cause expulsive osteolysis [5,6].

A definitive diagnosis can be made by histopathological examination. The diagnosis is difficult using fine-needle aspiration cytology alone, but can be made with a core needle biopsy, as in our case. However, this may be especially difficult in head and neck regions with a high risk of bleeding; thus, the indication should be judged carefully. Histopathological findings are characterized by a "patternless pattern", in which round to spindle-shaped cells are irregularly arranged, and a "staghorn", in which capillaries are branched. Immunohistochemical staining is positive for CD34, CD99, vimentin, and Bcl2, and negative for desmin, S100, and SMA [4-7]. Recently, STAT6 has been reported to be specifically positive for SFT [8].

The main treatment for SFT is surgical resection. Complete resection is important because a positive surgical margin is a poor prognostic factor [4]. However, it may be especially difficult to secure a sufficient margin in head and neck regions. Radiation therapy may be considered as an additional treatment for positive surgical margins, and as the primary treatment for recurrent or unresectable SFTs [9].

The prognosis for extrapleural SFTs is 4.3% for local recurrence and 5.4% for distant metastasis [10]. Positive surgical margins, age \geq 55 years, tumor size \geq 15 cm, number of cell divisions, and tumor necrosis are prognostic factors [3].

In the current case, we had to consider that the patient was a young woman who hoped for a cosmetically less invasive surgery, and determine how to completely resect a tumor against the anatomical complexity. To approach the skull base, we considered various skin incisions, including a lower lip midline incision, Weber-Ferguson incision, facial dismasking flap, mid-facial degloving, coronary incision, and cervical incision. With facial incisions (e.g. lower lip midline, Weber-Ferguson), there is a loss of esthetic outcomes. Although mid-facial degloving does not require a facial incision, a poor surgical field was predicted in our case. Therefore, a hemifacial dismasking flap was used.

The facial dismasking flap is useful approach for various regions, including the infratemporal fossa. An advantage of this technique is that the upper two-thirds of the maxillofacial region can be unfolded, preserving the facial nerve without any noticeable facial wounds. We consider this surgical approach as useful against tumors that widely occupy the infratemporal fossa [11].

Additionally, we considered various facial bone transection methods, including the zygomatic approach, orbito-zygomatic approach, lateral rhinotomy, facial translocation, partial maxillary swing approach, Le Fort I osteotomy, and mandibular swing approach. Fisch reported on an infratemporal fossa approach from the temporal bone [12]. Due to recent advances in endoscopic treatment, a transnasal approach using nasal endoscopy has also been reported [13]. In the current case, the temporal fossa was

widely occupied by the tumor, and no infiltration of the paranasal sinuses, nasopharynx, or temporal bone was observed. We selected a zygomatic approach because the visual field could be secured by adding a temporary transection of the zygomatic arch. This approach enabled the lateral pterygoid and temporal muscle, which comprised the surgical margins of the tumor, to be easily reached, and rendered it possible to resect the tumor completely.

In conclusion, in this case of an SFT in the temporal and infratemporal fossa, it was possible to achieve both complete resection and postoperative function using an appropriate surgical approach.

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