Salvage preoperative embolization of an infratemporal solitary fibrous tumor

A case report with review of the literature

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Abstract
Rationale: Head and Neck Solitary fibrous tumors (SFT) are very rare. They could be misdiagnosed as hemangiopericytomas (HPC).

Patient concerns: We report a 60 y o lady presenting with sinonasal mass, causing recurrent profuse bleeding.

Diagnoses: Hemangiopericytomas versus SFT were among the differentials, according to Radiological studies. Upon Biopsy, the diagnosis of SFT has been adopted.

Interventions: Salvage pre-operative embolization resulted in bleeding control, bridging the patient to surgery.

Outcomes: Post-operative course was uneventful, and patient symptoms resolved.

Lessons: This is the first case report of a sinonasal SFT, where pre-operative embolization has been employed as a salvage procedure. This treatment modality is promising, since it controls bleeding, bridges patient to surgery and decreases blood loss during the surgical procedure.

Abbreviations: HPC = hemangiopericytoma, SFT = solitary fibrous tumor, VEGF = vascular endothelial growth factors.

Keywords: embolization, hemangiopericytomas, solitary fibrous tumor

1. Introduction

Solitary fibrous tumors (SFTs) can occur anywhere in the human body, from the head[1] to the ischioanal fossa.[2,3] These spindle cell tumors were first reported in the pleura in 1931.[3] A similar tumor, hemangiopericytoma (HPC) was described in 1942 as a distinctive neoplasm showing characteristic histologic features, including the “staghorn” branching vascular pattern. Over the years, it has become apparent that HPCs and SFTs share considerable histologic and immunohistochemical features and thus, in the latest WHO classification of head and neck tumors, tumors previously classified as HPC are now classified as SFT.[4,21]

Head and neck SFTs are rare, and infratemporal ones are even rarer. Preoperative diagnosis is difficult and, only in few cases, paraclinical investigations have led to accurate diagnosis.[1,4] However, CD34 expression and positive nuclear STAT6 on immunohistochemistry are features that aid in diagnosis.[5,8]

Preoperative embolization has been reported in few cases. SFT occurring in the sinonasal compartments, act in a benign fashion, and only few patients present with malignant features.[8]

2. Case report

A 60-year-old female patient, presented with a 1-year history of minor recurrent epistaxis, and obstructive nasal symptoms. A fiberoptic nasal endoscopy showed a left nasal cavity mass originating from the maxillary sinus. The mass was brownish in color; it was soft, friable, and did not bleed with manipulation. Further investigation with a computed tomography (CT) scan showed a left nasal cavity mass involving the maxillary sinus, pterygopalatine fossa, and infratemporal fossa (Fig. 1A and B). A transnasal biopsy was diagnostic of SFT, and prompted profuse hemorrhage. Anterior nasal packing failed to control bleeding, and patient required transfusion of 2 blood units. The decision was to proceed with salvage arterial embolization.

2.1. Embolization Technique

Under local anesthesia with sedation, the right femoral artery was accessed. A sheath was placed in the left common carotid artery, and a 4Fr catheter was advanced in the external carotid artery. Angiographic evaluation of the tumor was obtained (Fig. 2A). A progreat 2.7Fr microcatheter was placed in the left internal
maxillary artery, and embolization was initiated with spherical 100 to 300 μm embospheres, followed by 300 to 500 μm polyvinyl alcohol. Then, the distal internal maxillary artery was occluded with a 6 mm × 15 cm concerto coil (Fig. 2B and C). Small residual arterial branches remained patent due to cross collaterals.

Complete and safe resection of the tumor was achieved by performing a maxillectomy with a lateral rhinotomy, using a Weber Ferguson approach, 10 days after the embolization. The postoperative period was uneventful, and the patient was discharged on the 6th postoperative day. Now the patient is on 4 months of follow-up postsurgery; she has complete resolution of her symptoms.

The final pathology again was diagnostic of SFT. The tumor demonstrated a cellular proliferation of bland spindle cells with a low mitotic count and no evidence of necrosis. Immunohistochemical studies showed that the tumor cells were positive for CD34. (Fig. 3A and B).

It is noteworthy mentioning that patient’s consent was obtained before publishing her case. An ethical committee approval is not required when considering publication of case reports, as per rules and regulations of the hospital ethical committee.

3. Discussion

Preoperative diagnosis of SFT remains a difficult task, although some specific diagnostic features on magnetic resonance imaging have been suggested. Complete surgical excision is usually curative. However, more aggressive behavior may be related to the tumor size (>15 cm), mitotic count (>4/10 hpf), presence of necrosis, and patient age (>55 years).
Radiotherapy should be reserved for incomplete resections, or for tumors showing malignant features. SFTs occurring in the sinonasal compartments are extremely rare, and act in a benign fashion. In a review of 20 SFTs, none of the nasal cavity tumors showed increased mitotic activity. About 12 cases of paranasal SFT have been published in the last 10 years (Table 1). All cases showed good outcome following surgical resection. Preoperative embolization has been previously described for patients with HPCs. Hanak et al have demonstrated decreased intraoperative blood loss, following preoperative embolization for patients with intracranial HPC. In this article, preoperative embolization resulted in better outcomes, because necrotic softening facilitates surgical resection. Similarly, this technique showed successful results for spinal, mediastinal, and orbital HPCs. In about 4 articles, preoperative embolization has been used for sinonasal HPC. In almost all cases, the maxillary artery was targeted, and none of the authors mentioned a single morbidity case. Some articles stated that preoperative necrosis might result in the release of a large amount of vascular endothelial growth factors (VEGFs), increasing the rate of lymphatic metastases, when bridging patients to orthotopic liver transplantation. However, it seems that lymphatic metastasis is regulated by other factors in addition to VEGF. Furthermore, time to surgery varies considerably between patients awaiting orthotopic liver transplantation and patient undergoing surgical resection few days after embolization. Despite these promising individual results, the role of preoperative embolization in sinonasal HPC and SFT is yet to be elucidated.

4. Conclusion
To the best of our knowledge, this is the first case of sinonasal SFT, where preoperative embolization has been employed as a salvage procedure. This treatment modality plays a major role in preoperative diagnosis, results in tumor necrotic softening, and reduces the risk of intraoperative bleeding. Although the role of preoperative embolization is not well defined; however, further studies should evaluate its indication as routine before the excision of SFTs.

Author contributions
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Table 1
Nasal and paranasal solitary fibrous tumors reported during the last 10 years.

| Age/sex | Size of the tumor | Location (s) | Treatment | Follow-up period in months/ (status of the patient) |
|---------|-----------------|--------------|-----------|-----------------------------------------------|
| Freiser et al (2014) | 38/F | 40 mm | Infratemporal | Surgery/transnasal approach | 6 (FOD) |
| Janjua et al (2011) | 33/F | N/A | Paranasal sinuses | Endoscopic surgery | 26 (FOD) |
| 36/M | N/A | Paranasal sinuses | N/A | 35 (FOD) |
| 41/M | N/A | Paranasal sinuses | N/A | 40 (FOD) |
| Kao et al (2016) | 55/M | 25 mm | Nasal cavity | N/A | 23 (FOD) |
| 46/M | N/A | Nasal cavity | N/A | LOF |
| 56/M | N/A | Nasal cavity | N/A | 21 (FOD) |
| 46/M | 42 mm | Paranasal sinus | N/A | 34 (FOD) |
| 47 | 27 mm | Paranasal sinus | N/A | 6 (FOD) |
| 16 mm | Paranasal sinus | N/A | 34 (DUC) |
| 37/M | 25 mm | Paranasal sinus | N/A | 7 (FOD) |
| Coca-pelaz et al (2011) | 48/M | N/A | Petrous bone (infratemporal aspect) | Surgery+radiotherapy | 24 (FOD) |

DUC = dead of unknown cause, F = female, FOD = free of disease, LOF = lost to follow-up, M = male, N/A = not available.
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Supervision: C. Tayar.
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Visualization: U. Hadi.
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Writing – review & editing: k. yammine, H.A. Nasser, v. Najjar.

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