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CASE REPORT

Massive Solitary Fibrous Tumor (SFT) of the infratemporal and pterygomaxillary fossa treated by combined endoscopic approach [version 1; peer review: 3 approved with reservations]

François Cloutier¹, Geneviève Lapointe², Sylvie Nadeau¹

¹Department of Ophthalmology and Otolaryngology - Head and Neck Surgery, Laval University, Hôpital de l'Enfant-Jésus, Quebec City, QC G1J1Z4, Canada

²Department of Neurological Sciences – Neurosurgery, Laval University, Hôpital de l'Enfant-Jésus, Quebec City, QC G1J1Z4, Canada

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Abstract

Solitary fibrous tumors (SFTs) were first described as spindle-cell tumors originating from the pleura. Until recently, there was some debate with regards to the name and origin of these tumors and the distinctions with the term haemangiopericytomas (HPCs), a rare type of vascular tumor.

Morphological, immunohistochemical and clinical features of HPCs were not specific for one entity. With the exception of myopericytoma, infantile myofibromatosis and HPC-like lesions of the sinonasal tract showing myoid differentiation, all other HPC-like lesions are best considered as subtypes of SFT. Due to their mesenchymal origin, we are now aware that SFTs may involve several extrapleural sites including soft tissues or meninges. When SFTs involve the skull base and show malignant histological characteristics, they can be an important challenge for the surgical team.

We report a case of a 54-year-old man complaining of poor vision and facial pain that had worsened over the last year. Computed tomography and magnetic resonance imaging indicated a large mass, involving the right infratemporal and pterygomaxillary fossa. A biopsy proved positive for a solitary fibrous tumor with malignant features. Surgery was performed using a combined approach of frontotemporal craniotomy and nasal endoscopy. The subtotal resection, conducted due to nodularity on the dura mater, was a success and was complemented by postoperative radiation therapy. Follow-up MRIs showed no recurrence of the tumor. To the authors' knowledge, this is the first reported case of a massive SFT involving this region, treated with minimal invasive surgery without any facial osteotomies.

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Approval Status ? ? ?

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version 1	?	?	?
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1. Masafumi Fukuda , Niigata University, Niigata City, Japan 2. Giulio Cantu , Istituto Nazionale Tumori, Milan, Italy 3. Rakesh K Chandra , Northwestern University Feinberg School of Medicine, Chicago, IL, USA			
Any reports and responses or comments on the article can be found at the end of the article.			

Keywords

Solitary Fibrous Tumor, Haemangiopericytoma, pterygomaxillary and infratemporal fossa, neoplasms

Corresponding author: François Cloutier (francois.cloutier.10@ulaval.ca)

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Introduction

Solitary fibrous tumors (SFTs) were first described by Klempere and Rabin⁷ in 1931 as spindle-cell tumors originating from the pleura. Until recently, there was some debate with regards to the name and origin of these tumors¹¹. The term haemangiopericytomas (HPCs) was used by Stout and Murray¹⁰ in 1942 when they described a type of vascular tumor derived from the capillary pericytes, previously described by Zimmermann in 1923.

However, over the years, the use of the term HPC has raised many issues. Morphological, immunohistochemical and clinical features of HPCs are not specific for one entity^{3,8}. With the exception of myopericytoma, infantile myofibromatosis and HPC-like lesions of the sinonasal tract showing myoid differentiation, all other HPC-like lesions are best considered as subtypes of SFT³. Due to their mesenchymal origin⁵, we now are aware that SFTs may involve several extrapleural sites including soft tissues or meninges^{5,8}.

Only a few cases of SFT have been described in the literature involving the skull base^{9,14}, but a massive malignant SFT of the soft tissue involving the infratemporal and the pterygomaxillary fossa is extremely rare and represents a multidisciplinary clinical and surgical challenge. The vast extent of the tumor and its localisation near vital structures makes it an important challenge for the surgical team, especially when the tumor shows malignant histological characteristics. Classical transfacial surgical approaches may leave unsightly scars with high morbidity. However, a combined approach with nasoendoscopy provides excellent access for tumor resection without any concerns over the aesthetic result.

To our knowledge, this is the first case reported of a massive SFT involving this region, treated with minimal invasive surgery without any facial osteotomies.

Case report

Written informed consent for publication of clinical details and clinical images was obtained from the patient.

History

Early December 2010, a 54-year-old caucasian man was referred to our otolaryngologist head and neck surgery department. He presented no relevant personal or family history. He had noticed progressive loss of vision and facial pain over the last year. The ophthalmologist, who saw him for decreased vision in the right eye, found a mass on the axial CT that was causing his compressive optic neuropathy.

Examination

The clinical examination revealed subtle proptosis of the right eye. Using flexible nasolaryngoscopy, we saw small bulging in the nasal cavity on the right side. All cranial nerves were intact and no lymph node enlargement was found. The patient was not complaining of epistaxis or nasal obstruction.

The first computer tomography scan with intravenous iodine contrast, requested by the ophthalmologist, indicated a lesion in the right pterygomaxillary fossa eroding the base of the skull to the temporal lobe and to the right cavernous sinus. There was also an extension in the orbital apex and erosion of the pterygoid process

with bulging of the posterior wall of the right nostril. The mass was visible with contrast gadolinium enhancement on the MRI. Its dimensions were 4.6cm anteroposterior × 3.3cm transverse × 4.7cm cranio-caudal (Figure 1–Figure 3).

Figure 1. Preoperative computed tomography of right pterygomaxillary fossa solitary fibrous tumor showing compression of the right optic nerve.

Figure 2. Magnetic resonance scan of the rhinopharynx - Global homogeneous enhancement with gadolinium of the mass.

Figure 3. Magnetic resonance scan of the rhinopharynx post-operatively showing subtotal removal of the tumor.

Shortly after the MRI, a transmaxillary endoscopic biopsy was performed. The result of the frozen tissue examination was compatible with a diagnosis of neuroendocrine carcinoma or hemangiopericytoma. The final pathology indicated a solitary fibrous tumor and this was confirmed by an expert otolaryngologist pathologist in Toronto. There was hypercellularity and a mitotic index of 4 mitoses per field, a sign of a highly aggressive tumor⁸ (Figure 4).

Treatment

The case was discussed at the multidisciplinary oncology clinic of our otolaryngology department. With neurosurgical and radio-oncology advice, it was decided to schedule surgery. A preoperative angiography was made with embolization of the tumoral branches by the right internal maxillary artery. It was made the day before surgery to minimize the risks of severe intraoperative bleeding.

The surgery began with the removal of the inferior and superior right turbinate. After that, we removed the median portion of the maxillary sinus. The tumor was bulging at this location and increased at the level of the sphenoid sinus.

Figure 4. A, B: Haematoxylin and eosin stain exhibiting a dense arrangement of polygonal cells with highly cellular patternless sheets with branching vascular pattern, thickened endothelial spindle cells with protuberant nuclei, C: CD34 staining consistent with SFT diagnosis, D: Bcl-2 staining intensely positive at magnification 20x.

To attain greater exposure, a Caldwell-Luc incision was made with minimal osteotomy of the anterior maxillary sinus. Using endoscopy, we witnessed the fibrous tumor covering the complete posterior wall of the maxillary sinus.

Laterally, the resection was made until we saw the fatty tissue of the infratemporal fossa. The vidian nerve had to be removed.

We excised the tumor with the capsule as much as possible. We had to terminate the surgery with a subtotal resection (STR), because it was noted that some nodularity persisted on the dura mater on the lateral-superior side of the tumor (Figure 3a,b). It was left there, because the sealing at this location would make it very difficult to reach and remove. In addition, it was considered a benign tumor with aggressive potential. As a result, radiotherapy was determined to be necessary, in any case.

The neurosurgeon performed frontotemporal craniotomy with right infratemporal fossa exploration. The debulking was made using ultrasonic suction-irrigation (Cavitron).

Postoperative course

One month later, the patient started external beam radiation (60 Gy by 30 fractions) of the paranasal sinus with infra temporal fossa and pterygomaxillary fossa.

On monthly follow-up, it appeared that the scars were healing well. There were no problems with the sinuses and no further complaints of facial pain. However, there is no improvement of the visual acuity in the patient's right eye. Subsequent MRI follow-up showed a continual small asymptomatic residual tumor, but no signs of recurrence more than one year later.

Discussion

SFTs are rare spindle-cell neoplasms of variable histological grades. Despite some confusion in the past regarding HPC and SFT, the relationship between the two is now more obvious. It is also known that SFT of the soft tissue can occur in the head and neck region and throughout the body^{4,6,12}.

Most cases of soft tissue SFTs occur in the early fifth decade of life⁸ with no sex predilection^{2,3}. Its occurrence is less than 2% of all soft tissue tumors⁸.

Skull base SFTs may include a wide variety of symptoms, although they are usually asymptomatic on presentation¹³. The symptoms manifest most frequently as a slowly expanding painless mass. Decreased vision, nasal obstruction, local pain, recurrent sinusitis, epistaxis, headaches, dural erosion, cerebrospinal fluid rhinorrhea, anosmia and lower cranial nerve palsies may all be part of the patient symptomatology.

SFTs from any site are usually benign and surgical resection alone is curative^{4,11}. However, malignancy is possible, although the criteria

remain imprecise. It may be suspected with the radiology exam (mass >10cm) or by the presence of metastasis¹³. The presence of infiltrative margins with surrounding tissues, high mitotic count (≥ 4 mitoses per 10 high-power high fields) of cellular pleomorphism and tumor necrosis also suggests malignancy^{4,8}.

The main treatment is surgical for benign and malignant SFTs. There is little evidence (principally from case reports) from adjuvant radiotherapy and none supporting the use of chemotherapy. But when histopathology suggests malignancy or when there are positive surgical resection margins, radiotherapy must be discussed, as for other sarcomas^{1,8}.

In conclusion, we successfully managed a case of massive solitary fibrous tumor of the soft tissue, involving the infratemporal and pterygomaxillary fossa. With the combination of conventional frontotemporal craniotomy with sinus surgery endoscopy, we were capable of removing most of the tumor while preserving vital structures close to the tumor and without the need for facial osteotomies, leaving the patient without any undesirable scars. With the addition of adjuvant radiotherapy treatment, we were able to achieve no sign of recurrence at the one-year follow-up.

SFT is very uncommon but should be part of the differential diagnosis in patients with a skull base lesion. Diagnosis is difficult but pathologists should be aware of the classical finding of this disease consisting of spindled cells in a disorganized pattern, with alternating hypocellular and hypercellular areas separated by hyalinized collagen and branching HPC-like vessels. Also the immunophenotyping staining positive for the presence of CD34 and Bcl-2 can be useful. The surgical approach needs to weigh risks and benefits of subtotal vs. total resection because of the surrounding vital structures.

Author contributions

François Cloutier: first author, acquisition of data, study design, participated in drafting the article and revising it critically for important intellectual content.

Gave final approval of the version to be submitted.

Geneviève Lapointe: Second author, substantial contribution to conception and design of this study.

Participated in drafting the article and revising it critically for important intellectual content, gave final approval of the version to be submitted and any revised version.

Sylvie Nadeau: Third author, substantial contribution to conception and design, participated in drafting the article and revising it critically for important intellectual content, and gave final approval of the version to be submitted and any revised version.

Competing interests

No competing interests were disclosed.

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Rakesh K Chandra

Department of Otolaryngology - Head and Neck Surgery, Northwestern University Feinberg School of Medicine, Chicago, IL, USA

This is an interesting case that is well illustrated and has some instructive elements. My only criticism is that it is not clear why a craniotomy was done if this was a benign (albeit aggressive) tumor and a subtotal resection followed by XRT was done anyhow. Perhaps the authors could discuss their surgical decision-making in this regard. The surgical defect in the postoperative MRI also appears to involve areas that were not previously involved radiologically.

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 10 March 2014

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Giulio Cantu

Cranio-Maxillo-Facial Surgery Department, Istituto Nazionale Tumori, Milan, Italy

The author write that the nasal endoscopic approach is better than the classical transfacial approach because this later may leave "unsightly scars with high morbidity". The emphasis in this sentence is probably excessive. The scars may sometimes be "unsightly" (although a good skin

closure makes the scar quite invisible) but I never saw “high morbidity” for the scar *per se*.

The authors write that the inferior portion of the tumor was capsulated whilst it was not the case of the portion adherent to the dura. The reported MRI images confirm this data. So, I believe that this tumor was resectable with the only infratemporal intracranial approach. Moreover, I believe that it was indicated a dural resection in order to achieve a complete radicality. I agree that the repair of the dura in that region is not easy but it is possible without CSF leakage (very rare in my experience, especially filling the dead space with the rotation of the temporal muscle or with a free flap). It is worthy of consideration that the postoperative radiotherapy is not a guarantee of success in case of macroscopic residual tumor, mainly when one is dealing with low grade tumors that often have a low radiosensitivity.

The reported sentence “With the addition of adjuvant radiotherapy treatment, we were able to achieve no sign of recurrence at one-year follow-up” has a very low oncologic meaning. I have seen relapses of solitary fibrous tumor after 12 years of the resection.

Finally, the postoperative figure 3A and 3B show a very enlarged liquor space: Why?

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 13 January 2014

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Masafumi Fukuda

Brain Research Institute, Niigata University, Niigata City, Japan

This is a rare case report of solitary fibrous tumor (SFT) occurred in the infratemporal and pterygomaxillary fossa treated by combined approach of frontotemporal craniotomy and nasal endoscopy. However, the points that the authors emphasized are not obvious. Do the authors want to stress the regions occurring the tumors, pathological findings, or the surgical approach? In the discussion, only the general considerations of SFT were described. Are there differences between the previous reports and the case regarding initial symptoms, radiological findings, surgical approach, or final outcome? The authors should show the tables including the information previously reported about SFT in the skull base.

Minor revisions

1. The authors used “minimum invasive surgery” in some parts. I think that craniotomy in addition to endonasal approach was not minimally invasive. The term “combined surgery”

or “combined approach” is better.

2. In the 4th paragraph of the introduction, the authors state “without any facial osteotomies”. However, in the 3rd paragraph of “Treatment”, the authors described that “a Cadwell-Luc incision was made with minimal osteotomy the anterior maxillary sinus.” Are these inconsistent?
3. In “Treatments”, the authors should describe the findings with respect to tumor characteristics, such as soft or hard, massive bleeding or not, massive invasion to the adjacent bone structures or not.
4. In the discussion, the first paragraph the authors have already said in the introduction. This paragraph should be deleted.
5. In the 6th paragraph of the discussion, the authors said that “With the addition of adjuvant radiotherapy treatment, we were able to achieve no sign of recurrence at the one-year follow-up”. SFT has slow growth rate and long-term follow-up is required, such as 10 or 20 years. The sentence should be deleted or re-wrote.

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

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