Solitary fibrous tumour of the cheek: Immunohistological diagnosis and radiosurgical therapy

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Abstract
Solitary fibrous tumour (SFT) is an uncommon spindle-cell neoplasm of mesenchymal origin occurring in the face.

We report a clinical case of SFT of the cheek. This entity is rarely observed on this site. The clinical behaviour is widely different and the diagnosis depends on the immunohistochemical results. In our report, we clarify these features. The treatment consists of complete surgical removal, better performed when previously associated with embolisation.

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1. Introduction

Solitary fibrous tumour (SFT) is an uncommon spindle-cell neoplasm of mesenchymal origin. It frequently appears in the pleura that was the first report in 1931.1

SFT arises from the ubiquitous interstitial stem cells in various soft tissues of the whole body, including the head. The cheek is rarely reported in the literature.2

The clinical behaviour is widely different and the diagnosis depends on the immunohistochemical examination.3 In our report, we clarify these features and emphasize on the great benefit of the embolisation when realized before surgery.

2. Case report

A 28-year-old woman presented with a swelling in the right cheek growing gradually over 4 months after her delivery. There was no history of trauma. On clinical examination, there
was a submucosal painless mass involving the vestibule of the right cheek. This lump was 4 centimetre (cm) in diameter, firm and relatively fixed on the deep plane. Both overlying skin and mucosa were intact. There was no trouble on the sensory and motor function of the face. Our patient had not any lymph node.

Radiologically, the patient was investigated with magnetic resonance imaging (MRI) with gadolinium contrast. The lesion was reported as a hypervascular solid mass within the fat of the right cheek (Fig 1).

In order to exclude bone involvement a computerised tomography (CT) study was performed. The CT did not indicate any bony destruction of the maxilla.

We suspected either a benign hypervascular tumour or a vascular malformation. That is why we decided to perform an embolisation before surgery to avoid an important bleeding.

Angiography indicated that the lesion is hypervascular and received arterial supply from muscular branches of the internal maxillary artery (IMA) and from the angular branch of the facial artery (Fig 2). Embolisation of these branches was achieved with absorbable gelatine sponge. Post embolisation angiogram showed complete devascularisation of the lesion (Fig. 3).

This woman was operated under general anaesthesia, one day after embolisation, with a vestibular incision discovering the tumour. It was not encapsulated but well circumscribed (Fig. 4). There was no lesion on the maxillary nerve (V2) and no erosion on the adjacent bone. The tumour was completely removed with no haemorrhagic accident.

The histological appearance of the specimen was that of a cellular fibrous tissue, within the spindle shaped fibroblastic cells were arranged in a rather “patternless” way. Immunohistochemical staining for markers CD34, CD99 and Bcl-2 was positive, whereas staining for S100 and neurofilament was negative. So SFT was confirmed.

Clinical examination and Post contrast MRI was performed one year after surgical resection and did not show any recurrence of the lesion.
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3. Discussion

Solitary fibrous tumour (SFT) is a rare benign soft-tissue neoplasm usually encountered in the pleura and was believed to arise from submesothelial primitive mesenchymal cells. Several extrapleural sites are described such as the respiratory tract, lung, breast, meninges and the cervicofacial region, even observed in the sinusual tract, the thyroid, the salivary glands, the tongue, the scalp and the cheek. Since then studies emphasize that SFT is distinct from pleural mesothelioma and the origin of SFT is the ubiquitous interstitial stem cells in various soft tissues.

In the oral region, SFT accounts for 3% of all the cases documented in the literature. Both men and women can be affected at the same range and mainly middle aged and elderly patients.

Symptoms depend on the site and the deepness of the tumour. Usually, slow growing and asymptomatic mass is described with an overlying normal skin and mucosa. There is no erosion of the adjacent bone as seen in our case. However, aggressive or malignant SFTs grow rapidly and can have a regional extension, even metastasis can occur to the lung, the liver and the bone.

Sometimes, few patients with SFT show a hypoglycaemia explained by the possibility of this tumour to secrete an “insulin like factor”.

Most cytogenetic studies report numerical abnormalities of chromosomes involving gain of chromosome 21, rearrangement of 9q22 and loss of chromosomes 3, 4, 9, 13 and 22.

These chromosomal changes may have a role of tumorigenesis of SFT but no genes were suggested or identified.

Radiologically, whether by magnetic resonance imaging MRI, computerized tomography CT or plain X-ray film SFT is not specific and appears as a richly vascularised soft tissue mass which is better identified with an angiography. Because of the well known hypervascularity of this tumour, embolisation of the blood vessels supplying the tumour is helpful before the surgical approach. We believe that surgery combined with radiological embolisation is the treatment of choice. We did not find this reported in the literature.

The histological diagnosis remains difficult because the wide morphologic spectrum exhibited by SFT generates a differential diagnosis essentially with hemangiopericytoma, leiomyosarcoma and desmoplastic fibroma.

The diagnosis of SFT is always and necessarily confirmed by an immunohistochemical analysis that shows the positivity for the immunoreactive CD34, CD99 and Bcl-2 of the tumour cells.

Both leiomyosarcoma and desmoplastic fibroma have no expression for CD34. However, we never find a positive expression for CD99 in hemangiopericytoma (HPC). At present, many authors believe that HPC and SFT represent the same pathologic entity.

It’s also important to search some histological features predicting malignancies such as the lack of circumscription, nuclear atypia, hypercellularity and the presence of necrosis. These features conclude to an aggressive or malignant SFT and even if the surgical treatment is complete it must sometimes be carried with radiotherapy and chemotherapy.

All the authors required a long term follow up, both clinical and radiological examination, to detect recurrence or its possible malignant transformation.

In conclusion, SFT is extremely rare in the cheek. Its diagnosis depends on immunohistological examination.

The treatment of choice is the complete surgical excision better performed when previously associated with embolisation. A long term follow up is necessary to detect recurrence or a possible malignant transformation.

Conflict of interest statement

None.

References