Solitary fibrous tumour of the tongue of an adolescent – a case report with immunohistochemical studies

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Solitary fibrous tumour (SFT) is a rare, spindle-cell, mesenchymal neoplasm with haemangiopericytoma-like branching vascular pattern. The tumour has been reported in various locations but the oral cavity is a distinctly uncommon region. We describe a case of solitary fibrous tumour (SFT) in the tongue of a 15-year-old girl. Microscopically it had features characteristic for SFT and a common immunohistochemical profile but uncommonly increased mitotic count.

Introduction

Solitary fibrous tumour (SFT) is a rare, spindle-cell, mesenchymal tumour with prominent haemangiopericytoma-like branching vascular pattern, which is commonly observed in middle-aged adults [1]. Initially, SFT was described in the pleura [2] but recently it has been reported in various locations, including: soft tissue of extremities, abdominal cavity, mediastinum, thoracic wall, retroperitoneum, meninges, spinal cord, lungs, liver, gastro-intestinal tract, salivary glands, thyroid, nasal and oral cavity, etc. [1]. SFT may be found at any location, but the oral cavity is a distinctly uncommon location with only 3 cases formerly described in the tongue [3-5] and to our knowledge SFT has not been previously reported in the tongue in adolescents.

Case report

A 15-year-old girl was admitted to our hospital for the presence of painless nodule of the tongue, which had appeared a few weeks earlier. Under general anaesthesia the lesion was excised.

On gross examination the tumour was described as a well-circumscribed, uncapsulated, tan-white mass, which had a diameter of 1 cm. The cut surface appeared solid.

Microscopically the tumour consisted of patternless, haphazard, hypercellular proliferation of spindle cells. Cellular areas separated from each other by thick bands of hyalinized collagen and branching haemangiopericytoma-like vessels. Non-atypical, spindle-shaped cells with scant cytoplasm had dispersed chromatin with vesicular nuclei (Fig. 1). The tumour had numerous mitoses (Fig. 2, arrow) – seven per ten high-power fields (7/10 HPF) (Fig. 2). There was no necrosis, cytological atypia or infiltrative margins but the tumour was excised incompletely.

Tumour cells stained strongly positive for CD34 (Fig. 3) and there was strong and diffuse positivity for bcl-2 oncoprotein (Fig. 4). The tumour cells were focally positive for S-100 and negative for desmin. A diagnosis of solitary fibrous tumour was made.

Discussion

Solitary fibrous tumour is an uncommon, fibroblastic neoplasm which was first described in the pleura by Klemperer and Rabin in 1931 [2]. But recently extrapleural SFTs have been increasingly reported and now they are even more common than pleural lesions [6-9]. The oral cavity is a distinctly uncommon site and

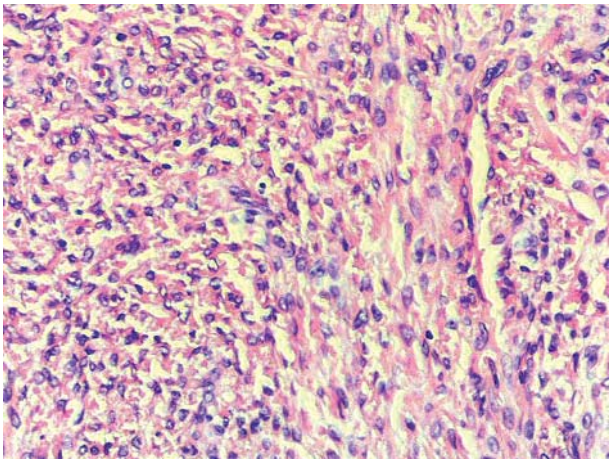

Fig. 1. SFT – Microscopically the tumour consisted of patternless, haphazard, hypercellular proliferation. Cellular areas separated from each other by thick bands of hyalinized collagen and branching haemangiopericytoma-like vessels (HE 200 ×)

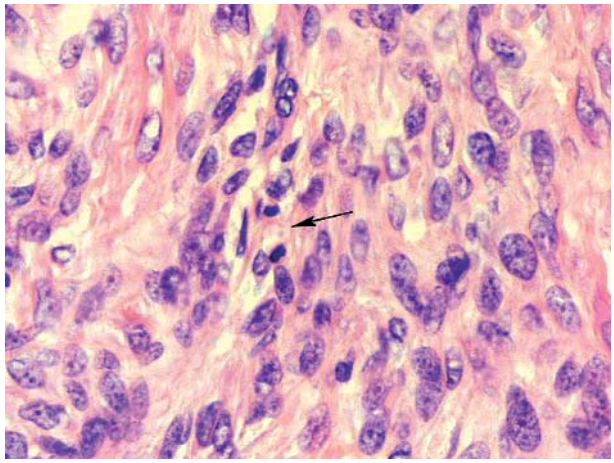

Fig. 2. Tumour had numerous mitoses – seven per ten high-power fields (7/10 HPF) (HE 400 ×)

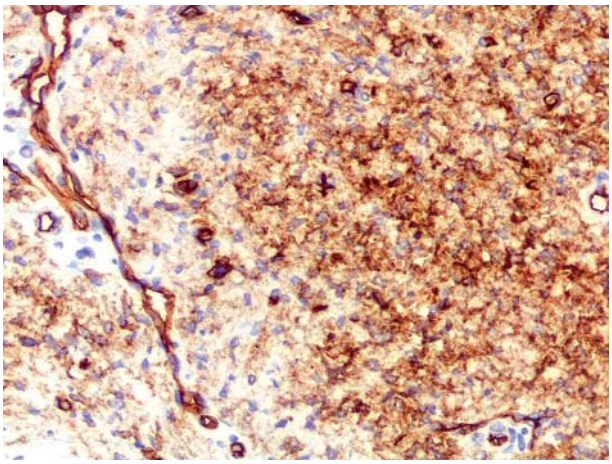

Fig. 3. Tumour cells stained strongly positive for CD34

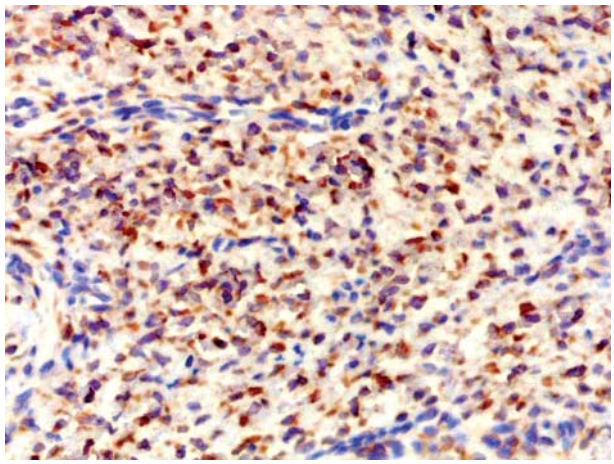

Fig. 4. Strong and diffuse positivity for Bcl-2 in tumour cells

most cases have reported involvement of the buccal mucosa [10, 11]. In the tongue, SFT is extremely rare, with only a few cases previously described [3-5], and to our knowledge SFT has not been previously reported in the tongue in adolescents. Clinically solitary fibrous tumour is usually a slow-growing, painless mass in middle-aged adults, rarely in adolescents, with no sex predilection [1, 7]. Our patient was 15-year-old girl with a painless tongue nodule, which is uncommon at this age and location. Microscopically, SFTs are often described as tumours with patternless growth because they present a wide range of morphological features. Mitoses are generally scarce and rarely exceed 3 mitoses per 10 high-power fields [1, 3, 5, 7, 9]. In our case the tumour had more mitoses (7 per 10 high-power fields), which could have suggested malignant character of the lesion. Criteria of malignancy for SFT include: large tumour size (> 50 mm), disseminated disease at presentation, infiltrative margins, high cellularity, nuclear pleomorphism, areas of tumour necrosis and an increased mitotic index (> 4 mitoses per 10 HPF) [6, 7, 9]. Our case presented only two of these features of malignancy: high cellularity and an increased mitotic index; therefore we qualified our tumour as a benign lesion.

Although for some authors the characteristic immunohistochemical profile for SFT is positive for vimentin, CD34, Bcl-2, and negative for desmin, SMA, S100 and cytokeratins [3, 5, 6, 10], WHO has stated that tumour cells of SFT are immunoreactive for CD34 and CD99, variably positive for EMA, Bcl-2, SMA, and of focal and limited reactivity for S100 protein, cytokeratins and desmin [1]. The immunoreactivity of our case was strongly positive for CD34 and bcl-2 and also focally positive for S100, which is not very

common for SFT but according to WHO [1] and some authors [8, 9] possible.

CD34 is a helpful marker for distinguishing SFTs from other soft tissue tumours, especially with another marker, which is strongly immunoreactive positive in SFTs, Bcl-2 protein [4-6, 9, 12]. Suster *et al.* [12], who examined over 50 cases of SFTs and their reactivity for CD34 and Bcl-2, noticed that CD34 has positive reactivity in the majority of cases and Bcl-2 has strong, diffuse reactivity in every examined case [12]. So, co-expression of positive reactivity with CD34 and Bcl-2 strongly supports a diagnosis of SFT. But evidently we must remember that not only SFTs are CD34 and Bcl-2 positive, spindle-shaped neoplasms. The others are: haemangiopericytomas, dermatofibrosarcomas protuberans, gastro-intestinal stromal tumours, neural tumours and smooth muscle tumours [12]. According to this knowledge our interpretation of immunoreactivity with these two markers must be determined in appropriate clinical course, by means of histopathological features and other immunohistochemical markers such as vimentin and CD99.

In conclusion, we have described a case of SFT in the tongue of a 15-year-old girl. In this age and location the tumour is very rare. Microscopically it had characteristic SFT features and common immunohistochemical profile but uncommonly increased mitotic count. On the basis of all performed examinations, the diagnosis of solitary fibrous tumour of the tongue was established. Because the diagnosis was made only a few months ago we do not have any follow-up yet or any information about eventual recurrence.

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