Solitary fibrous tumour of the buccal mucosa: morphological and histopathological differences and similarities in relation to other spindle cell neoplasms

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Abstract
Introduction
This case report describes an unusual tumour of the oral and head neck region—the solitary fibrous tumour. 

Case report
A patient is reported with a relatively fast growing lump in the right cheek. It was initially diagnosed as a dermofibrosarcoma and a secondary operation was performed with extended excision. The final diagnosis was later changed to solitary fibrous tumour by the pathologist. The histopathological features of the solitary fibrous tumour and resemblance with other spindle cell tumours are discussed.

Conclusion
Despite difficulties in diagnosis, the clinician and pathologists must be aware of this tumour especially in oral soft tissue which can lead to incorrect treatment planning and higher morbidity.

Introduction
Solitary fibrous tumour (SFT) is a rare, mesenchymal neoplasm most commonly originating from mesothelial-lined surfaces such as the pleura and peritoneum. The diagnosis of SFT is primarily histological and is difficult due to the fact that many other spindle cell neoplasms present similar features. Publication frequency of intraoral tumours with the diagnosis SFT has been increasing during the past decade, partially due to its relatively new recognition as an intraoral lesion and reclassification of previously filed cases of spindle cell tumours. Thus far, there have been only a few reports of malignant SFT involving the oral cavity. In any event, awareness of this lesion is important in the differential diagnosis of oral soft tissue tumours. This paper reports a case of SFT of the buccal mucosa.

Case report
A 54-year-old male was referred to the Department of Oral and Maxillofacial Surgery, Sahlgrenska Academy, University of Gothenburg, for examination and treatment of a growth in the buccal mucosa. The past medical history was unremarkable. The patient first noticed a ‘bulge’ on the inside of the right cheek about a year earlier but it is possible that he had had it for an even longer period. For the last 6 months, he had felt some discomfort due to interference with the cheek mucosa when chewing.

Extra-oral examination revealed neither asymmetry nor swelling of the face. Intra-orally the cheek mucosa with an area of 15 mm diameter and an irregular reddish-yellow surface was revealed. On palpation a well-defined, freely movable, firm mass. On dissection, a spherical and well defined, almost capsulated-like duct that was adjacent to the tumour was revealed. The postoperative course was uneventful with good healing. The initial histopathological diagnosis was Dermatofibrosarcoma protuberans (DFSP), which is another rare lesion in the oral cavity. DFSP is a low grade malignancy and therefore a second intervention was performed swiftly since the margins of the excised sample was microscopically not free from tumour tissue. An extended excision was performed under general anaesthesia with margins of at least 10 mm, but with consideration taken to preservation of important structures, aesthetics and function. Even this time the postoperative course was uneventful.

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Microscopic examination

Haematoxylin and eosin-stained sections from the first sample revealed a well-defined, non-capsulated, soft tissue tumour directly approximating the overlying epithelium. Most of the tumour tissue was not only rich in collagen but also myxoid areas were seen. In the deep portion invasion of glands and ducts was seen (Figure 1). High magnification showed spindle-shaped and ovoid cells without significant cytologic atypia (Figure 2). Mast cells and lymphocytes were moderately present. Mitotic figures were very sparse and neither xanthomatous cells, fat cells nor giant cells were found. Immunohistochemistry showed intense reactivity for the cell proliferation marker Ki-67 was recorded only in a few cells (Figure 3d). The initial diagnosis, Dermatofibrosarcoma protuberans, was revised to SFT at the final assessment. Analysis of the second sample gave similar findings and the margins were free from tumour tissue.

Discussion

Fibroblastic tumours and tumour-like proliferations in the oral cavity show an extremely varied microscopic appearance and present a diagnostic challenge. CD34 reactivity is seen in several spindle cell lesions and does not alone allow a definitive diagnosis of SFT5–13. Thus, CD34 is expressed in tumours of neural origin, smooth muscle tumours, haemangiopericytoma (HPC), dermatofibrosarcoma (DFS), spindle cell lipoma (SCL) and benign fibrohistiocytoma (FHC). In nodular fascitis, however, CD34 activity was lacking14,15. In the differential diagnosis versus SFT, immunostaining for S-100 can exclude tumours of neural origin since these stain positive and while SFT does not. Smooth muscle tumours and nodular fascitis show lesional reactivity for smooth muscle actin while SFT only shows reactivity in vessel walls.

Abundant mast cells are seen in SFT and not in HPC16. However, an associated inflammatory cell infiltrate that included mast cells and eosinophils was noted in the majority of HPCs16. In SFT, reactivity for smooth muscle actin is found in the vessel walls while it is not in HPC and furthermore, the CD34 reactivity in HPC is restricted to endothelial cells while in SFT, also the tumoural cells show reactivity13.

The present case did not show a prominent storiform pattern and xanthomatous cells were absent which excluded the possibility of FHC and DFS. Furthermore, hyalinization was noticed in the present tumour and this is not a feature of DFS. Mast cells have been demonstrated also in solitary and multiple DFS, in particular in the layers between the DFS lesion and the overlying epidermis3.

Spindle cell lipoma was initially considered a possibility but was excluded because no mature fat cells could be demonstrated.

Correct diagnosis is a prerequisite to quick and adequate treatment, and more importantly, as in this case, in the planning of resection margins. In this case, despite initial cytology tests, no malignancy was suspected and therefore, the decision was to excision without margin. The second extended excision was based on the initial diagnosis DFSP and was swiftly performed in order to minimise morbidity associated with malignant lesions. Subsequent analysis revealed that the histomorphological patterns of the resected
samples fitted more with the diagnosis SFT. The rareness of SFT in the oral soft tissues and infrequent exposure of pathologists to such soft tissue spindle cell neoplasms coupled with overlapping histological patterns can make diagnosis difficult. One of the first reports made within the past decade, of intraoral SFT was of Suster et al.17 However, as pointed out by Ide et al.,18 this lesion has been described earlier under other names.

Conclusion

Despite difficulties in diagnosis, the clinician and pathologists must be aware of this tumour especially in order to avoid misdiagnosis with other more aggressive neoplasms of oral soft tissue which can lead to incorrect treatment planning and higher morbidity.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Abbreviations list

DFS, dermatofibrosarcoma; DFSP, Dermatofibrosarcoma protuberos; FHC, fibrohistiocytoma; HPC, haemangiopericytoma; SFT, Solitary fibrous tumour; SCL, spindle cell lipoma

References

12. Weiss SW, Nickoloff BJ. CD-34 is expressed by a distinctive cell population in peripheral nerve, nerve sheath tumors

Figure 2: Collagenous tumour tissue with bland spindle-shaped and ovoid cells without atypia (haematoxylin-eosin stain ×400).
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Figure 3: (a) Immunohistochemical staining for CD34 showing intense reactivity in the tumour tissue (×50). (b) Immunohistochemical staining for S-100 protein. Immunoreactivity in gland structures while the tumour cells don’t show any reactivity (×150). (c) Immunoreactivity for smooth muscle specific actin in vessel walls. The tumour cells are negative (×150). (d) Immunoreactivity for Ki-67 only in a few scattered cells (×150).