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Short communication

Oral nodular fasciitis: Report of a case of the buccal mucosa

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ABSTRACT

Nodular fasciitis is a benign, reactive, proliferative spindle-cell lesion, usually located at the subcutaneous tissues or muscle fascia. Clinically, it manifests as a soft-tissue mass with well-defined margins and fixed to the adjacent structures. Because of its rapid growth rate, rich cellularity and relatively high mitotic activity, nodular fasciitis is sometimes misdiagnosed as a sarcoma. Accurate diagnosis is based only on histopathological examination. A rare case of nodular fasciitis of the buccal mucosa in a 50-year-old female patient is presented.

1. Introduction

Nodular fasciitis is defined by the World Health Organization as a benign proliferative fibroblastic lesion (Martínez-Blanco et al., 2002). It was first described by Konwaler et al. (1955) and is also known as pseudosarcomatous fasciitis, pseudosarcomatous fibromatosis, proliferative fasciitis and infiltrative fasciitis (Haddad et al., 2001; Sharma et al., 2008).

Although the pathogenesis of this entity remains unknown, it appears that it is a self-limiting lesion, reactive or inflammatory in nature, rather than a true neoplasm. Nodular fasciitis is often mistaken for a sarcoma due to its rapid growth, rich cellularity and relatively high mitotic activity (Han et al., 2006). Therefore, proper diagnosis can be achieved only after histopathological examination (Martínez-Blanco et al., 2002; Nair et al., 2004).

Nodular fasciitis is a relatively uncommon lesion, which most often affects the trunk and upper limbs. In 7–20% of the reported cases, the lesion is located at the head and neck region. Intraoral location of the lesion is very unusual (Sharma et al., 2008).

Clinically, lesions are described as a well-circumscribed mass of soft or elastic consistency, which adheres firmly to underlying tissues and structures. They are generally asymptomatic, although sensitivity or pain in the region may be described in some cases (Shlomi et al., 1994). Lesions are most commonly found in the subcutaneous tissue, muscle fascia and muscle (Lenyoun et al., 2008).

Treatment of choice is complete surgical removal while recurrence is uncommon (Martínez-Blanco et al., 2002; Lenyoun et al., 2008).

2. Case report

A 50-year-old white woman presented to a private dental clinic for evaluation of a painless swelling to the right buccal mucosa. According to the patient, the swelling had been present for 8 months, and there was no history of trauma or inflammation in the area. The patient reported that she used to rub with her hand the skin at the site of the lesion. Her medical history was non-contributory.

Examination revealed a circumscribed soft-tissue mass of the right buccal mucosa, located respectively to the premolar region of the mandible. The lesion measured approximately 1.5 cm in diameter, covered by normal mucosa (Fig. 1). The mass was moderately firm on palpation and non-tender. A panoramic radiograph revealed no pathology, while ultrasound examination showed the presence of a hypoechoic tumorous mass in the right cheek, without the presence of any hemorrhagic elements within the lesion (Fig. 2).

Under local anesthesia the lesion was resected by an intraoral approach. It was dissected off the adjacent muscle fibers of the buccinator muscle and was removed *in toto*. The surgical site was closed primarily. The postoperative course of the patient was uneventful.

The histological examination revealed a relatively well-demarcated but not encapsulated mass with moderate cellularity, composed of interlacing fascicles of monomorphic spindle cells with uniform elongated oval nuclei and eosinophilic cytoplasm. The described cells were arranged in a myxoid stroma. Chronic

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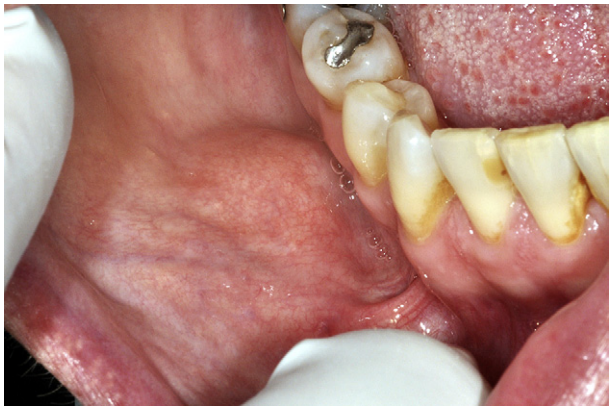

Fig. 1. Clinical view of the lesion.

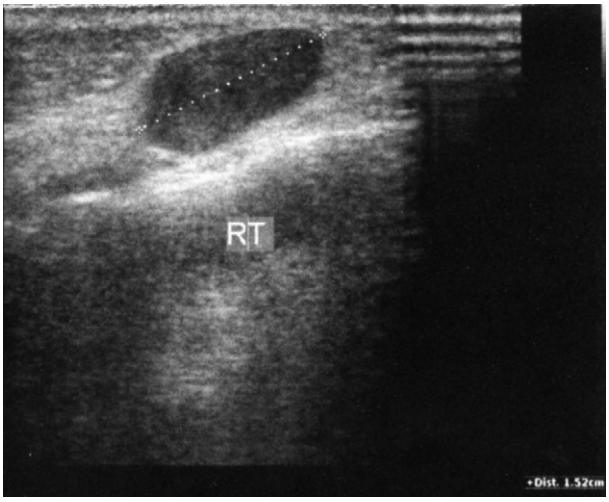

Fig. 2. Ultrasound examination showing the presence of a hypoechoic tumorous mass.

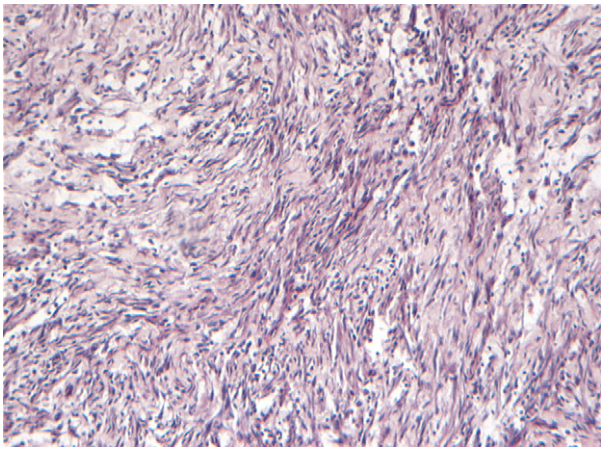

Fig. 3. Interlacing fascicles of monomorphic spindle cells with uniform elongated oval nuclei and eosinophilic cytoplasm, arranged in a myxoid stroma (hematoxylin and eosin, original magnification ×10).

inflammatory cells were scattered throughout (Fig. 3). Immunohistochemically, the cells were reactive with antibodies against smooth muscle actin and vimentin, but showed no reactivity with antibodies against S-100 protein, desmin, parakeratin, CD34 and CD68 (Fig. 4).

The cell proliferation index Ki67 was <5%, an immunophenotype which revealed that the cells were of fibroblastic and/or myofibroblastic origin. The morphological and immunohistochemically findings were consistent with the diagnosis of nodular fasciitis.

One year after the excision the patient was disease free without signs of recurrence.

3. Discussion

Nodular fasciitis affects mainly people aged 20–40 years. Males and females are equally affected. It usually presents as a rapidly growing soft-tissue mass in the upper extremities and the trunk, but it may occur in any location. A 7–20% head and neck predilection is recorded, usually appearing in the skin of the face, the parotid sheath and the subcutaneous tissues overlying the mandible and zygoma. The intraoral location of nodular fasciitis is rare and it may affect the tongue, the floor of the mouth, the alveolar mucosa and the buccal mucosa (Martínez-Blanco et al., 2002; Sharma et al., 2008).

Nodular fasciitis of the buccal mucosa is an uncommon lesion. Reviewing the English literature for cases of orofacial nodular fasciitis between the years of 1994 and 2005, Han et al. (2006) have found only 8 well-documented cases of nodular fasciitis of the buccal mucosa. In the same article they reported one more case, in a 48 years old female. The female to male ratio was 8:1, with a mean age of 40 years. Up to 2009 no other similar cases of nodular fasciitis of the buccal mucosa have been reported (Table 1).

The pathogenesis of nodular fasciitis remains unknown. It seems that it is a self-limiting lesion, reactive or inflammatory in nature rather than a true neoplasm. Trauma may be a causative factor. However, injury preceding the appearance of the lesion is seldom reported. The clinical picture of nodular fasciitis is quite worrying, as it appears as a rapidly growing mass with a diameter that is usually no more than 4 cm. Nodular fasciitis arises from the muscle fascia and extends into the subcutaneous tissues or the underlying muscle. The lesion typically is well-circumscribed, non-encapsulated and firmly adherent to the adjacent structures (Haddad et al., 2001; Nair et al., 2004).

Because of its rapid growth, presence of spindle-shaped cells, high mitotic activity, rich cellularity, and due to the fact that the lesion may spread along peritoneal spaces, nodular fasciitis is often misdiagnosed as a sarcoma or other malignant mesenchymal neoplasms. However, nodular fasciitis is a benign lesion that never metastasizes (Han et al., 2006). In contrast, soft-tissue sarcomas of the head and neck aggressively invade surrounding tissues and disseminate haematogenously (Kim et al., 2009).

The clinical and imaging features of nodular fasciitis are not pathognomonic. Histopathological examination and immunohistochemistry are mandatory for the precise diagnosis. The spindle cells of nodular fasciitis contain vimentin and variably actin, but do not contain desmin, keratin or S-100 protein (Eversole et al., 1999; Haddad et al., 2001; Han et al., 2006).

The differential diagnosis includes other tumors of mesenchymal origin which are characterized by the presence of spindle cells, such as myxoid neurofibroma, myxoid dermatofibrosarcoma, fibrosarcoma and leiomyosarcoma (Martínez-Blanco et al., 2002; Han et al., 2006).

Treatment of choice for nodular fasciitis is complete conservative surgical excision. However, the lesion is often self-limiting. Yanagisawa and Okada (2008) report a case of nodular fasciitis of the cheek that degenerated and spontaneously regressed after biopsy. The authors conclude that “nodular fasciitis with myxoid histology can change to that with fibrous appearance gradually with time, thus bringing about spontaneous regression”. However, the mechanism of regression is unclarified at present. The

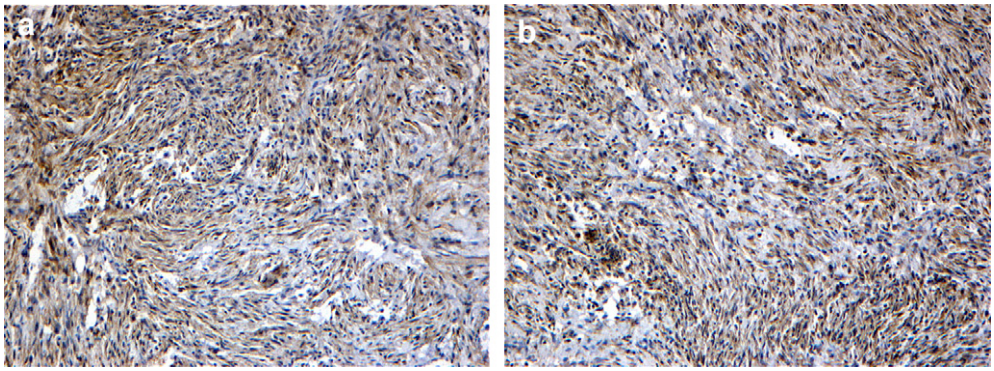

Fig. 4. Immunohistochemical markers for the lesion (original magnification ×10): a. SMA (+), b. Vimentin (+).

Table 1
Clinical data of documented cases of intraoral nodular fasciitis of the buccal mucosa, published from 1994 to 2009.

Reference	Age/gender	Duration of lesion	Size* (cm)	Signs and symptoms	Follow-up†
Badia et al. (1994)	76/F	6 months	1.8	N/M	1.5 years
Shlomi et al. (1994)	30/F	2 months	2.0	Discomfort	6 months
Alkan et al. (2001)	35/F	4 months	1.7	Painless	1.25 years
Nair et al. (2004)	37/F	2 months	1.5	Painless	1.5 years
Dayan et al. (2005)	50/F	2 months	2.0	Asymptomatic	4 years
	43/F	3 weeks	2.0	Asymptomatic	1 year
	42/F	2 weeks	0.8	Tenderness	2 years
	38/M	N/M	N/M	Asymptomatic	5 years
Han et al. (2006)	48/F	1 month	2.0	Asymptomatic	9 years
Present case	50/F	8 months	1.5	Painless	1 year

M: male; F: female; N/M: not mentioned.
* Greatest diameter.
† No recurrence in any of the cases.

prognosis is favorable. Recurrence is uncommon (1–2% of all cases of oral nodular fasciitis) and if it happens, occurs shortly after surgical removal, probably due to incomplete resection (Haddad et al., 2001; Han et al., 2006).

4. Conclusion

A well-documented case of oral nodular fasciitis, occurring at the buccal mucosa, is presented. Because of the rarity of the lesion and non-specific clinical and imaging features, special caution is imperative in order to obtain accurate diagnosis and appropriate treatment. Attention and increased index of suspicion are always required by Clinicians and Histopathologists and nodular fasciitis, although somewhat rare, should be listed in the differential diagnosis of oral soft-tissue tumorous masses.

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