Nodular Fasciitis of the Oral Cavity with Partial Spontaneous Regression (Nodular Fasciitis)

Marina Lara de Carli · Karin Sá Fernandes ·
Décio dos Santos Pinto Jr. · Andrea Lusvarghi Witzel ·
Marília Trierweiler Martins

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Abstract Nodular fasciitis is a lesion found in the subcutaneous fascia that microscopically presents as a benign proliferation of fibroblasts and myofibroblasts, which may be mistaken for a sarcoma due to clinically rapid growth. Diagnosis is by histopathology and of the immunohistochemical profile. We describe a case of nodular fasciitis in the oral cavity that demonstrated partial spontaneous regression. The patient was a 32-year-old man with a buccal mucosal mass, which had grown rapidly for 45 days. On microscopic examination, the lesion displayed a well-delineated but not encapsulated proliferation of spindle cells, with a nodular growth pattern. Immunohistochemical analysis showed positivity of the spindle cells for the antibodies against smooth muscle actin and muscle-specific actin (HHF-35). Treatment of such lesions typically involves complete conservative excision, but the lesion may regress eventually in the absence of definitive treatment.

Keywords Nodular fasciitis · Oral cavity · Reactive lesion · Spindle cells · Spontaneous regression

Introduction

Nodular fasciitis (NF) is a benign, idiopathic, reactive proliferation of myofibroblasts found in the subcutaneous fascia and presents as a rapidly growing mass, within a duration of 1 month or less [1], leading to it frequently being mistaken for a sarcoma [2]. It was first described by Konwaler et al. [3] as subcutaneous pseudosarcomatous fibromatosis. Although somewhat rare, it should be considered in the differential diagnosis of oral soft-tissue tumor masses [4], including sarcomatoid carcinoma, fibrosarcoma, leiomyosarcoma, and neurofibroma [2]. The pathogenesis of NF remains unknown although trauma may be a causative factor [4, 5]. The clinical course in patients with NF is entirely benign. Complete local excision, with as much tissue-sparing as possible is considered to be the treatment of choice [2, 4, 6, 7]. We present here an unusual case of a NF of the oral cavity that showed partial spontaneous regression.

Case Report

A 32-year-old white male was seen at the Oral Medicine Clinic of the School of Dentistry with the complaint of painless swelling in the left buccal mucosa, which he had first noticed 45 days before. The patient reported that the lesion had grown rapidly in the last 10 days. Intraoral examination revealed a 2.0-cm painless firm, nodular mass with defined borders and covered by reddish mucosa with no bleeding (Fig. 1).

M. L. de Carli · K. Sá Fernandes
Department of Stomatology, School of Dentistry,
Universidade de São Paulo, São Paulo, Brazil

K. Sá Fernandes (✉)
Disciplina de Patologia Bucal, Faculdade de Odontologia,
Universidade de São Paulo, Av. Professor Lineu Prestes,
2227, São Paulo, SP 05508-000, Brazil
e-mail: karinsf@gmail.com

D. dos Santos Pinto Jr. · M. T. Martins
Department of Stomatology, Oral Pathology, School
of Dentistry, Universidade de São Paulo, São Paulo, Brazil

A. L. Witzel
Department of Stomatology, Oral Medicine, School of Dentistry,
Universidade de São Paulo, São Paulo, Brazil

Fig. 1 Clinical aspect of the lesion as nodule in the buccal mucosa

The differential clinical diagnoses included salivary gland tumor and pyogenic granuloma. An incisional biopsy was performed, and the specimen was sent to the Surgical Oral Pathology Service for histologic examination. The specimen consisted of a single brownish soft-tissue fragment with an elastic consistency, measuring $20 \times 5 \times 4$ mm.

Upon microscope examination, the lesion displayed a well-delineated but not encapsulated proliferation of spindle cells with a nodular growth pattern. The cells were organized in intersecting bundles and sometimes in a storiform fashion. Areas with a myxoid aspect were scattered throughout the sections. The cells were large with uniform elongated oval nuclei, eosinophilic cytoplasm and evident nucleoli. Mild pleomorphism and rare typical mitotic figures were seen scattered in the field. In the limited depth of the lesion, bundles of skeletal muscle in close contact with the spindle-cell proliferation could be seen. There was a mild chronic inflammatory infiltrate dispersed throughout the lesion. Immunohistochemistry showed positivity of the spindle cells for antibodies against smooth muscle actin and muscle-specific actin (HHF-35) but was negative for antibodies against S-100 protein. The Ki-67 antibody was used to determine the proliferation index, and typically less than 5 % of the cells were positive (Fig. 2). After immunohistochemical analysis and histopathologic examination, a diagnosis of nodular fasciitis was made.

Because the histologic diagnosis was nodular fasciitis and the nodule appeared to exhibit a tendency to resolve spontaneously, a wait-and-see policy seemed to be justified [8]. At the patient's 1-month follow-up, the mass had clearly regressed and measured 1.5 cm in diameter (Fig. 3). The lesion continued to reduce in size and a follow-up examination at 2 months revealed that the mass was 0.4 cm in size with a fibrous appearance (Fig. 4). This was excised surgically. Three months after the excision, the patient was disease free without signs of recurrence.

Discussion

The NF is a rare lesion that affects people between the third and fifth decades of life [9], with an approximately equal sex distribution. The reported duration of growth ranges from 3 weeks to 2 years, and the reported size ranges from 1.0 to 4.7 cm [9]. Anatomically, NF most commonly occurs on the upper extremities (48 %), followed by the trunk (20 %), the head and neck (15–20 %) and the lower extremities (15 %) [10]. NF in the orofacial region is rare, but when they do occur, they are most commonly located in the buccal mucosa, tongue, floor of the mouth or alveolar mucosa [5].

The most common histologic location of NF is in the subcutaneous tissue and fascia muscle. Intraoral nodular fasciitis is extremely rare; perhaps because fascia is not prominent in the oral cavity [11]. The most common location for oral lesions are places of origin or insertion of the masticatory muscles, which are vulnerable to trauma. In the present case, the clinical lesion was located in the buccal mucosa, probably in the fascia of the buccinator muscle, a region well suited to trauma from chewing, but the patient had reported no history of trauma to the area.

The pathogenesis of this entity remains unknown. Some authors believe that this lesion may be of traumatic, infectious or inflammatory origin [4, 12, 13]. The clinical features of NF in the oral maxillofacial region are similar to those in other regions. Clinically, lesions are described as a well-circumscribed mass of soft or elastic consistency, not encapsulated, which adheres firmly to underlying tissues and structures [4, 7, 11]. The lesion in this case report was similar to what is observed in the literature. It was small and solitary, had a benign reactive process, adhered firmly to the mucosa, was characterized by rapid growth, covered by ulcerated mucosa, and firm on palpation, with well-demarcated borders.

Due to the rapid growth of the lesion, presence of mitosis, spindle cells and rich cellularity, the differential diagnosis of sarcoma is important. However, NF is a benign lesion that never metastasizes [9]. In contrast, soft-tissue sarcomas of the head and neck aggressively invade surrounding tissues and disseminate hematogenously [14]. Furthermore, NF can show a tendency to resolve spontaneously after incisional biopsy, which is not seen in sarcomas [8]. NF can be differentiated from other facial mesenchymal tumors, including sarcomatoid carcinoma, fibrosarcoma, leiomyosarcoma, and neurofibroma, based on histology, molecular biology, and immunohistochemistry [2]. A characteristic immunohistochemical profile of NF include positive staining for vimentin, smooth muscle actin and muscle-specific actin, indicating a myofibroblastic differentiation. NF do not express keratin, S-100 protein, or desmin [15]. Nevertheless, the immunohistochemical profile is supportive but not pathognomonic for this lesion.

Fig. 2 Histopathologic and immunohistochemical features. **a** The lesion was represented by a well-delineated but not encapsulated proliferation of spindle cells inside the lamina propria (hematoxylin-eosin, 25 $\times$ in the original). **b** Higher magnification reveals the spindle-shaped cells have large oval nuclei and discrete pleomorphism

(hematoxylin-eosin, 400 $\times$ in the original). **c** This photomicrograph shows the close contact between the proliferation and the normal skeletal muscle seen in the deep margin (hematoxylin-eosin, 100 $\times$ in the original). **d** Positive immunohistochemical staining for smooth muscle actin (streptavidin–biotin, 100 $\times$ in the original)

Fig. 3 The lesion 1 month status-post incisional biopsy, showing signs of regression

Fig. 4 Two months status-post incisional biopsy, it is possible to notice significant regression of the lesion

Histologically, NF can be classified into three subtypes, according to the plane of tissue involved: subcutaneous, intramuscular and fascial. The lesion is usually attached to the fascia from which it arises and extends into the subcutaneous fat in an irregular fashion. Occasionally, it arises from the fibrous septa of the subcutaneous fat and only secondarily extends to the fascia [9]. The typical features include haphazardly arranged pleomorphic spindle cells

with uniform, elongated, oval nuclei and eosinophilic cytoplasm in a myxoid stroma, high mitotic activity, with no atypical forms present, and chronic inflammatory cells scattered throughout [4, 8, 9]. All cases diagnosed as NF show the same immunohistochemical marker profile. All exhibit variability in vimentin positivity. Smooth muscle actin was seen in fibroblastic cytoplasmic processes;

immunohistochemical staining was positive for HHF-35 and negative for S-100 protein [2, 16, 17], which corroborates with the present report that lesions were reactive with antibodies against smooth muscle actin and HHF-35 but showed no reactivity with antibodies against S-100 protein.

Treatment of NF typically involves complete conservative excision, and recurrence occurs rarely (in 1 to 2 % of all cases) [9, 11, 16], but many believe that any recurrence indicates error in the initial diagnosis [2]. However, because the lesion will not recur after excision and may spontaneously regress even if incompletely excised, further treatment is not always necessary after accurate diagnosis. NF with myxoid histology can degenerate into one with a fibrous appearance gradually over time. This case demonstrates that the components of a NF with myxoid histology can change with time, thus bringing about spontaneous regression, in agreement with other authors [8]. Because gene expression profiles of nodular fasciitis reveal the upregulation of genes encoding inflammatory cytokines and chemokines in comparison with other types of fibroblastic entities [18], one could speculate that these lesions are poised to regress, or that they are inflamed and would eventually regress in the absence of definitive treatment.

In the present case, although the lesion had shown some spontaneous regression, the patient underwent surgical re-excision. Three months after the excision, the patient was disease free without signs of recurrence. Accurate diagnosis is important for appropriate treatment and avoiding unnecessary surgical procedures.

Conflict of interest The authors declare no conflict of interest.

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