Nodular Fasciitis in the Oral Cavity

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Abstract

Rapidly growing soft-tissue lesions in the oral and maxillofacial region can represent a variety of diagnoses involving radically different treatment modalities. Accurate diagnosis is important to avoid unnecessary and often mutilating surgery. Nodular fasciitis is a rapidly proliferating fibroblastic lesion that presents as a tumour-like mass. Although up to 20% of cases occur in the head and neck region, lesions of the oral cavity are extremely rare. A case of oral nodular fasciitis is described, and a review of the literature is presented.

MeSH Key Words: *case report; fasciitis/pathology; mouth diseases/pathology*

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Nodular fasciitis, also known as nodular fibrositis, subcutaneous fibromatosis,[1] pseudosarcomatous fasciitis, proliferative fasciitis, subcutaneous pseudosarcomatous fibromatosis and infiltrative fasciitis,[2,3] is an uncommon tumour-like fibroblastic proliferation that was first reported by Konwaler and others in 1955.[4] It is a well-recognized, although an uncommon, fibroblastic growth that usually arises within the subcutaneous tissues of the extremities and trunk.[3-7] Approximately half of the reported cases occurred in the upper limb, and up to 20% of all cases occur in the head and neck region.[3,6,8] This lesion is important because it is often mistaken for sarcoma[5,9] due to its rapid growth, rich cellularity and mitotic activity.[1,3,4]

Case Report

A 9-year-old girl presented with a swelling of rapid onset in the upper right lip. The patient had severe cerebral palsy and was unaware of the lesion, which had been discovered by her parents. At the time of presentation, the swelling had been present for approximately 3 weeks. On examination, a mass measuring approximately 1 cm in diameter was palpable intraorally. The lesion was firm and very mobile and could be easily translated below the labial mucosa. There was no pain or irritation in the area. In addition to the cerebral palsy, the patient's medical history included severe seizure disorder and multiple admissions to hospital for pneumonia. She was taking a variety of medications, including anticonvulsives and bronchodilators.

With the patient under general anesthesia, the lesion was removed via a small incision through the labial mucosa (**Figs. 1,2**). The mass was easily dissected out and was submitted for histopathology (**Figs. 3,4**).

The specimen was a spindle-cell lesion composed of loose, plump, bipolar fibroblastic cells arranged in a storiform pattern with loose myxoid background in many areas. Extravasated red blood cells and a mixed inflammatory infiltrate were also present throughout the areas. Few mitotic figures were identified. Special staining for vimentin yielded positive results, but staining for actin, keratin, smooth muscle actin, S-100 and desmin yielded negative results. The final diagnosis was inflammatory nodular fasciitis.

Pathogenesis

The exact cause of nodular fasciitis is still unknown, but there is little doubt that it is the result of a self-limiting, reactive process rather than a true neoplasm. Most authors believe that the lesion represents some type of reactive or inflammatory condition triggered by local injury or infection.[2,3,6,9,10] Stout[11] noted that some of his patients with nodular fasciitis had a history of trauma preceding the appearance of the lesion. However, if prior injury plays a role, it is difficult to explain why trauma is reported in only 10% to 15% of cases and why the lesion is most common in the upper half of the body.[3] The term "fasciitis" implies that the lesion originates in the fascia and that it is of an

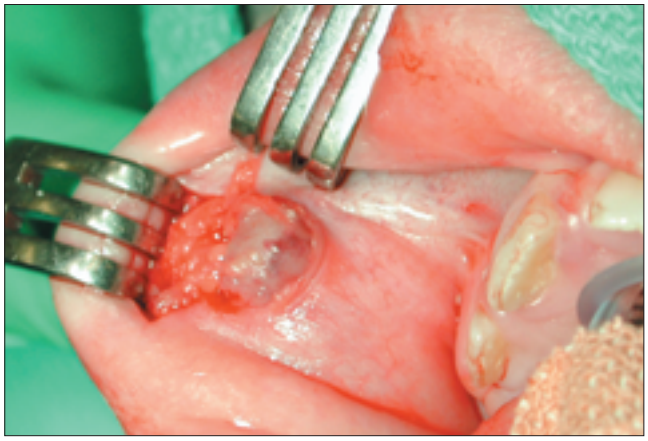

Figure 1: *Exposure of a rapidly growing intraoral lesion in a 9-year-old girl.*

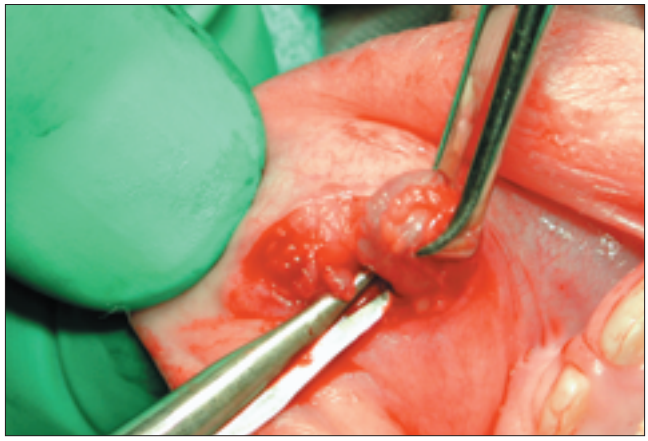

Figure 2: *Removal of the lesion.*

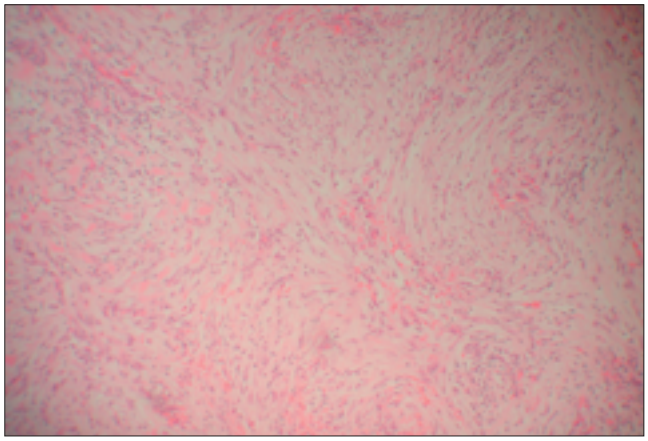

Figure 3: *Histopathological examination, using haematoxylin and eosin (H & E) stain, reveals the storiform pattern. (Original magnification × 100.)*

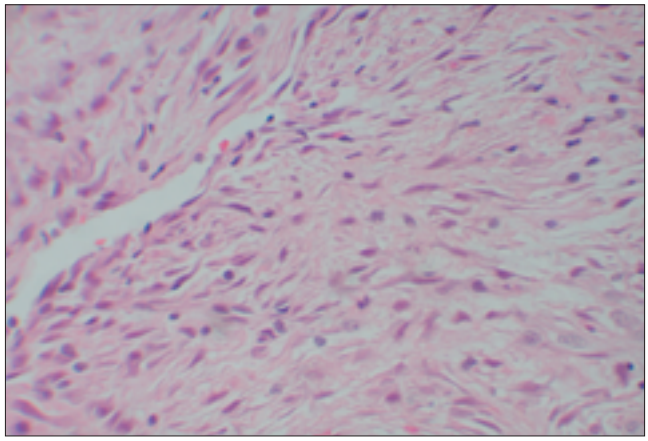

Figure 4: *Histopathological examination using H & E stain, in the centre of the nodule, showing spindle cells. (Original magnification × 400.)*

inflammatory nature, but neither of these aspects has been proven.[9] Although the oral cavity is subject to repeated trauma, intraoral nodular fasciitis is extremely rare, perhaps because fascia is not prominent in the oral cavity.[10] Many more such lesions in the oral cavity will have to be evaluated before trauma can be considered a valid etiologic factor.

Clinical Findings

Nodular fasciitis is most common in the third decade of life but occurs at all ages. Males and females are equally affected.[3,11-13] It usually presents as a rapidly growing soft-tissue mass[1-19] ranging from 0.4 to 10.5 cm in diameter[12] but usually not exceeding 4 cm.[1,6,17] It is circumscribed but not encapsulated, somewhat tender and fixed to the adjacent structures, but with freely movable overlying skin.[1,3,5,12,13] There is almost always a history of tenderness or pain, for a period of a few weeks to a few months, without evidence of systemic disease. Perineural extension, which may be responsible for the pain reported by some patients,[3,15] is sometimes observed. The macroscopic appearance is unreliable for diagnosis, since the lesion may be situated in subcutaneous, intramuscular or fascial tissues.[3,5,12,13]

Although nodular fasciitis may occur virtually anywhere in the body, there is a distinct predilection for certain sites; almost half of these lesions occur in the upper extremities, especially the volar aspect of the forearm. Next in frequency is the trunk, particularly the chest wall and back.

Nodular fasciitis of the head and neck is rare in adults but more common in children. In fact, the head and neck region is by far the most common single site in children. Nodular fasciitis is seldom encountered in the lower extremities, and only a few cases have been reported in the oral mucosa.[3]

The diagnosis of nodular fasciitis should be made more cautiously when such a lesion occurs in the head and neck area.[3,5,9] Because of the scarcity of this lesion in the head and neck region, most cases have been reported singly or in groups of 2 or 3 cases.[5,17] Nodular fasciitis has been reported to occur in the skin of the face,[5] the nose,[1] the

Table 1 **Differential diagnosis of spindle-cell tumours**

Benign lesions	Malignant lesions
Nodular fasciitis	Fibrosarcoma
Fibromatosis	Liposarcoma
Fibroma	Hemangiosarcoma
Neurofibroma	Rhabdomyosarcoma
Leiomyoma	Leiomyosarcoma
Neurilemoma	Neurosarcoma
Fibrous histiocytoma	Malignant fibrous histiocytoma
Myxoma	Myxosarcoma
Desmoid tumour	
Proliferative myositis	
Sclerosing angiomatosis	
Granulation tissue	

infraorbital area,[18] the parotid gland,[5] the mucosa opposite the apical portion of the upper front teeth,[13] the angle of the mandible,[17] the mandibular ridge,[10] the submental area,[1,6,12] the mucosa of the cheek,[5,8,10,15,17] the labial mucosa,[7] the mental foramen area and the tongue[5] and in association with the buccinator muscle.[14]

The clinical features of nodular fasciitis, with sudden onset of a rapidly growing tumour, may suggest an aggressive or even malignant process. Because of the sometimes alarming histologic characteristics, pathologists have frequently diagnosed the lesion as a malignancy. However, it is now generally agreed that nodular fasciitis is benign and never metastasizes.[3,6,9,10,17] A number of distinct spindle-cell tumours may be considered in the differential diagnosis of nodular fasciitis (**Table 1**).

Histological Findings

Price and others[1] classified nodular fasciitis according to 3 histological types: myxomatous, intermediate and fibromatous. The myxomatous type of nodule (type 1) was characterized by an abundant, almost acellular central stroma, with more abundant and often plump fibroblasts at the periphery. These nodules were also quite vascular, with parallel arrangement of the capillaries. The picture was rather suggestive of granulation or repair tissue. In the more cellular intermediate (type 2) nodules, more fibres were present, often oriented in bundles. Numerous slit-like spaces, often containing erythrocytes, were observed, along with scattered lymphocytes and macrophages. The fibromatous (type 3) nodules were similar to type 2 nodules but contained more collagen. The fibroblasts were shaped more like spindles, rather than being plump, and the fibres were often arranged in interlacing bundles. Well-formed capillaries and small veins, rather than slit-like vascular spaces, were seen. However, no correlation was found between the histologic type and the clinical features of the nodule.[1]

Stout[11] preferred the term "pseudosarcomatous fasciitis" because it emphasized the 2 important features of the lesion, namely, its simulation of the appearance of a sarcoma, even though it is not biologically malignant, and the presence of inflammatory stigmata in most cases.

The criteria reviewed here represent the most helpful features to distinguish nodular fasciitis from malignant spindle-cell neoplasm, especially fibrosarcoma. Fibrosarcoma does not usually manifest as a circumscribed tumour; its fascicles tend to be longer and more gently curved, and their occurrence is not associated with focal myxomatous areas.[5,12] Inflammatory infiltrate and extravasated red blood cells are absent, as are peripherally radiating capillaries. In nodular fasciitis, mitotic figures can be present in alarming numbers, but this feature alone should not eliminate fasciitis from the differential diagnosis. The presence of central necrosis or degeneration has not been mentioned in past publications, except by Price and others,[1] who recorded its occurrence in some of their type 1 cases of nodular fasciitis. Central necrosis or degeneration in nodular fasciitis has no apparent adverse implications.[5]

All cases diagnosed as nodular fasciitis have shown the same immunohistochemical marker profile. All exhibited some degree of positive reaction to vimentin staining, from weak to strong. Smooth muscle actin is seen in fibroblastic cytoplasmic processes, and staining for this compound is generally more intense in the more cellular regions.[18,19] Staining for S-100 is usually negative,[19] and the lack of staining for desmin, which is a more specific marker for muscle, also points to the fibrohistiocytic nature of the lesion.[8,18]

Discussion

The importance of recognizing the true nature of fasciitis relates to the potential for overtreatment on the basis of microscopic features that are suggestive of malignancy.[5,6,8,9,12,13,15] Ordinary light microscopy and immunohistochemical examinations are means of confirming the diagnosis definitively.[8,10] Nodular fasciitis is entirely benign and most likely a reactive lesion, with no metastatic tendency.[3,6,9,10,17] Given the experience of the case reported here, it should also be emphasized that the lesion may appear in the oral cavity and should consequently be considered in the differential diagnoses of all fibrous lesions of recent onset in this region. Nearly all examples of the lesion have been effectively treated by local excision. Recurrence, which occurs rarely[3,6,10] (in 1% to 2% of all cases) and then only soon after excision,[3] is probably the result of continued

growth after incomplete removal of the tissue. In cases of spontaneous regression, once the diagnosis has been made, no further treatment appears necessary.[1,3,4,16]

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