CASE REPORT

Nodular Fasciitis of the Orofacial Region: An Uncommon Differential

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Abstract Nodular fasciitis is a benign proliferative spindle-cell lesion that presents as a rapidly growing mass arising from the subcutaneous fascia, leading it to be erroneously diagnosed as a sarcoma. These lesions commonly present in individuals in their third to fifth decades of life with no definite gender predilection. They are frequently located on the extremities and the trunk and infrequently in the head and neck region. Lesions in the orofacial region are uncommon. We describe an interesting case of orofacial nodular fasciitis in a 21-year-old boy that essentially summarises the clinical features and management of this pseudosarcomatous reactive proliferative lesion. Clinicians and more so maxillofacial surgeons need to be aware of the distinctions between nodular fasciitis and its malignant mimic (sarcomas) in order to limit overtreatment and the resultant treatment-related morbidities.

Keywords Nodular *fasciitis* · Orofacial region · Sarcoma

Introduction

Nodular fasciitis is defined as a benign and reactive fibroblastic growth extending from the superficial fascia and onto the subcutaneous tissue or muscle (WHO definition). These tumors have no gender predilection and are more common in the third to fifth decades of life [1]. These lesions are frequently located on the extremities and the

trunk and infrequently in the head and neck region. Lesions in the orofacial region are uncommon [2]. We describe an interesting case of orofacial nodular fasciitis in a 21-year-old boy that essentially summarises the clinical features and management of pseudosarcomatous reactive proliferative lesion.

Case Report

A 21-year-old boy with no significant medical history or history of trauma presented to us with a recurrent lesion in the left cheek (left nasolabial region) which had been growing over the course of a month. He gave a history of having undergone an attempted excision of a similar swelling at another centre for a presumed diagnosis of a sebaceous cyst. The gritty intra-operative feel of the swelling and its possible adherence to the underlying premaxilla aroused a suspicion of a malignancy and the patient was hence referred to our centre.

Clinical examination at our centre revealed a residual 2 × 2 cm subcutaneous swelling in the left nasolabial region, the skin overlying the lesion showed the scar of the previous excision. General examination and examination of the rest of the head and neck was unremarkable. A review of the histopathology slides of the incisional biopsy at our centre revealed elongated and spindled cells seen as interlacing bundles and fascicles, seen with moderate cytoplasm and spindled nuclei. Mitosis varied from 0 to 1/high power field. Further immunohistochemistry revealed immunopositivity to vimentin and smooth muscle actin (SMA) and immunonegativity to Epithelial Membrane antigens (EMA) keratin and desmin. The histiocytoid cells showed immunopositivity to CD68. Less than 10 % of the cells showed positivity to ki-67. The histology

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suggested a diagnosis of Nodular *fasciitis* (Fig. 1a, b). An MRI of the head and neck region revealed the residual well-defined T2 hyper intense swelling measuring $1.8 \times 1.7 \times 1.4$ cm in the muscular plane of the left nasolabial region which showed homogenous enhancement with IV contrast. There was surrounding soft tissue edema, the underlying bone was however normal (Fig. 2).

The patient was taken up for a definitive re-excision of the residual lesion encompassing the scar of the previous excision (Fig. 3). The adjacent skin and sub cutaneous tissues were mobilised to achieve a primary closure (Fig. 4). The histopathology of the re-excision also showed features suggestive of nodular fasciitis with a cellular

subtype. The patient continues to be on regular follow up for close to 8 months post his definitive surgery.

Discussion

Konwaler et al. [3] was the first to describe nodular fasciitis in 1955 under the name of subcutaneous pseudosarcomatous fibromatosis. It has also been known by various other terminologies like nodular fibrositis, subcutaneous fibromatosis, pseudosarcomatous fasciitis and infiltrative fasciitis. Prior to its recognition as a distinct entity, these lesions were often misclassified as some form of sarcomas. The

Fig. 1 a, b H&E x10: Showing a lesion of Nodular fasciitis composed of elongated and spindled cells seen as interlacing bundles and fascicles, seen with moderate cytoplasm and spindled nuclei. Mitosis varied from 0 to 1/high power field

Fig. 2 An MRI of the head and neck region revealed the residual well-defined T2 hyper intense swelling measuring $1.8 \times 1.7 \times 1.4$ cm in the muscular plane of the left nasolabial region which showed homogenous enhancement with IV contrast

Fig. 3 **a** Clinical photograph on presentation at our centre. **b** Intra-operative photograph on excision of the lesion

Fig. 4 Immediate post-operative photograph following primary closure

exact aetiology of nodular fasciitis is by and large unknown and the role of trauma in initiating the lesion is also not clear [4]. It has however been largely believed that these lesions are triggered by local injury or an inflammatory process which causes an unusual proliferation of myofibroblasts (incidentally believed to be the cell of origin). Pregnancy is also believed to be a risk factor and this has been attributed with the associated hormone-related changes [5].

These lesions of nodular fasciitis are generally small, solitary and more common in the third to fifth decades of life with no definitive sex predilection. These lesions may be confused with other more common benign soft tissue

tumors such as fibroma, lipoma, desmoid tumor, chondroma, myxoma, schwannoma, atypical fibroxanthoma or rarely sarcomas [1]. Nodular fasciitis can at times be deceptively similar to a sarcoma by frequently extending along the fascial planes and exhibiting high cellularity, nuclear atypia and mitoses [6, 7]. However, they have been shown to be distinctively benign with no potential for malignant transformation. Nodular fasciitis can be further histologically classified into myxoid, cellular, and fibrous subtypes based on the amount and type of extracellular matrix. A relationship between these subtypes and the age of the lesion has been proposed i.e. the younger the lesion, the more the amount of the myxoid component.

The immunohistochemical profile for nodular fasciitis includes immunopositivity for smooth-muscle actin (SMA), vimentin and occasionally CD68 indicating a dual myofibroblastic and histiocytic differentiation. The lesions are usually immunonegative for keratin, S-100 protein and desmin. Histomorphologic features aided by an appropriate immunohistochemical profile are thus useful in the accurate diagnosis of nodular fasciitis [8].

Complete resection is the treatment of choice in all cases of nodular fasciitis [9]. There have however been a few reports suggesting that a partial excision may be acceptable as residual lesions are believed to resolve spontaneously and recurrences if any are rarely reported [10]. Watchful waiting and delayed surgery has hence been proposed as possible management options in lesions wherein the proliferation is slow or where resection would be difficult or cause significant mutilation.

In conclusion, our case demonstrates that, although infrequent in the orofacial region, nodular fasciitis should be considered as a possible differential diagnosis. It is essential that clinicians and more so maxillofacial surgeons are aware of the distinctions between nodular fasciitis and

its malignant mimic (sarcomas) in order to limit overtreatment and the resultant treatment-related morbidities.

Compliance with Ethical Standards

Conflict of interest None.

Ethical Issues All procedures performed in this case report were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Appropriate informed consents have been obtained.

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