Oronasal nodular fasciitis: a case report and literature review

Fasciíte nodular oronasal: relato de caso e revisão da literatura

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RESUMO

Introdução: A presença de uma massa solitária, firme, de rápido crescimento na região orofacial tem uma longa lista de diagnósticos diferenciais, que vão desde lesões benignas às mais fatais e malignas. Fasceíte nodular (FN) é uma proliferação reativa benigna do fibroblasto que emerge dos tecidos moles, provavelmente em resposta a uma lesão no local. Apesar de comumente vista nas extremidades superiores (50%), a FN orofacial é menos comum, com uma incidência inferior a 20%, afetando principalmente adultos na 4ª a 5ª décadas de vida. **Relato do Caso:** Neste relato, descrevemos um caso único de fasceíte nodular oronasal agravada por um trauma em uma criança de 6 anos do sexo feminino. De acordo com nosso conhecimento, este é o primeiro relato de caso de fasceíte nodular na literatura que surge nesta localização anatômica.

Descritores: Fasciite/patologia. Fasciite/cirurgia. Cavidade Nasal.

ABSTRACT

Background: The presence of a solitary, firm, progressively or rapidly growing mass in the orofacial region has a long list of differential diagnoses, ranging from benign lesions to more fatal malignant ones. Nodular fasciitis (NF) is a benign, reactive proliferation of fibroblasts that emerges from the soft tissues, most probably in response to a local injury. Although commonly seen in the upper extremities (50%), the orofacial NF is less common with an incidence less than 20%, affecting primarily adults in 4th to 5th decades of life. **Case Report:** In this report, we describe a unique case of oronasal nodular fasciitis aggravated by trauma in a 6-year old female child. To the best of our knowledge, this is the first case report of nodular fasciitis in the literature that arises from such an anatomic location.

Key words: Fasciitis/pathology. Fasciitis/surgery. Nasal Cavity.

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INTRODUCTION

Nodular fasciitis (NF) is uncommon progressive or rapid proliferation of fibroblasts that emerges from the soft tissues[1]. It was first reported by Konwaler et al.[2], in 1955, who described it as subcutaneous pseudosarcomatous fibromatosis. However, Price et al.[3] used the term nodular fasciitis, in 1961.

NF can affect any age group. But, it mostly occurs adults in their 4th to 5th decades of life[4-8]. There is no race or gender predilection. Moreover, it can occur in any part of human body with the exception of the viscera[9,10]. The most common sites of occurrence, in the order of decreasing frequency, are the upper extremities (48%), trunk (20%), head and neck (15-20%), and lower extremities (15%)[8,11,12]. In the orofacial region, NF affects adults in their fourth and fifth decades of life[1] more than children and young adolescents[7,13]. There is no gender or race predilection as well[1]. Clinically, it appears as a solitary, firm, well-indurated, mobile mass with a history of progressive or rapid growth over a short period of time[1,14], and are occasionally associated with a history of pain. NF can also appear as a solitary mass or in the form of subcutaneous plaque[15]. The solitary masses often originate from the subcutaneous tissue[13,16]. Other sites of origin include fascia[4,12], intramuscular[5,13,17,18], Submucosa[1,10,13,19,20], and intra-vascular[21-23]. The intravascular type is extremely rare, whereas the intra-muscular one is usually deceptive and may mimic malignant counterparts at clinical and radiographic evaluation due to its larger size, infiltrative nature, deeper locations, and ill-defined borders on radiographs. The histologic variants of orofacial NF can be arranged into three types: myxomatous (reactive), intermediate, and fibromatous (proliferative)[8,23]. The myxomatous type is the most common histologic picture in orofacial NF[1,23].

In this article, we present a case of trauma induced nodular fasciitis that arises from the right alar base with an extension into the oral and nasal cavities in a 6- year old female child. Furthermore, updates on epidemiologic data, clinical presentation, pathogenesis, diagnosis, and correlation between radiography and diagnosis of orofacial NF are discussed.

CASE REPORT

A 6-year-old healthy girl, of Asian-Indian decent, was referred to our facility for a second opinion regarding a rapidly growing mass of the right nostril. Initially, the patient complained of an asymptomatic, progressively growing lesion located at the right alar base. The lesion had been present for at least a month, according to the parents.

During examination, FNAB of the lesion was done, but the result was non-specific due to inadequate specimen. Within two weeks, the lesion tripled in size, and thus, the patient visited our facility for further examination. At the time of presentation, patient denied any history of allergic rhinitis, nasal congestion, headaches, epistaxis, anosmia, or sinusitis. The lesion appeared as subcutaneous mass, red-pinkish in color, tender to palpation, distorting the anatomy of right alar base, and associated with thinning of the overlying skin (Figure 1). The right nostril was almost blocked by the lesion. Intra-oral examination showed the presence of well-indurated, firm, non-ulcerated, mobile mass bulging into the oral cavity. CT scan revealed a well–circumscribed, homogenous mass that did not cause any destruction of the adjacent soft and hard tissues. The mass measured 3×3×2 cm and extended into both oral and nasal cavities (Figure 2).

Neurologic examination was unremarkable. Surgical excision of the lesion was performed under general anesthesia via trans-oral approach. There was a certain degree of difficulty in removing the entire mass as a single unit due to adherence of the mass to the surrounding tissues. Thus, it was removed in piecemeal. Postoperative healing was uneventful. The patient was followed up for more than 12 months and no recurrence was noted. The histology was consistent with myxomatous type of NF (Figure 3 a 5).

Figure 1 – Preoperative aspect.

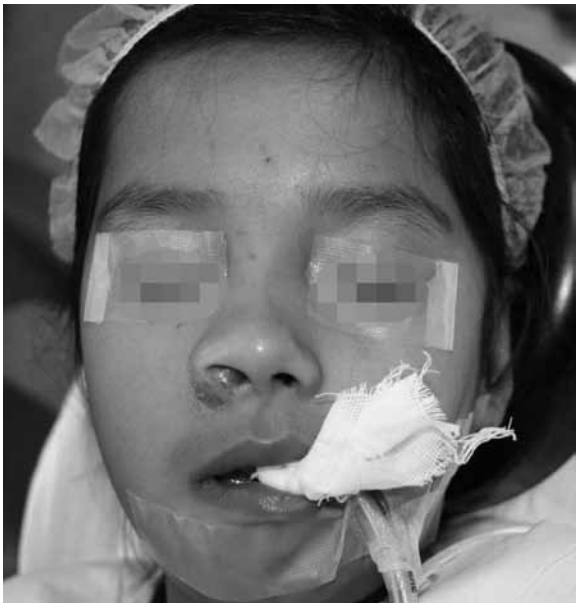


Figure 2 – CT scan revealed a well–circumscribed, homogenous mass extended into both oral and nasal cavities.

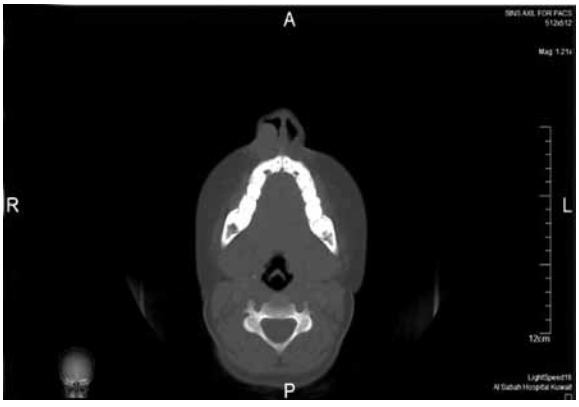


Figure 3 – Fascicles of spindle cells + sprinkling of lymphocytes.

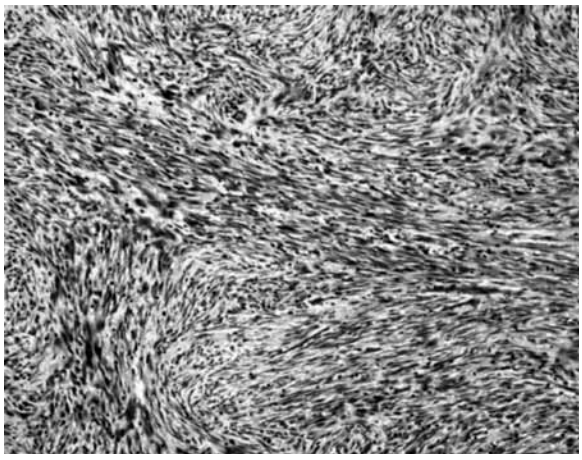


Figure 4 – Higher magnification: lymphocytes, red blood cells, mitosis (arrow).

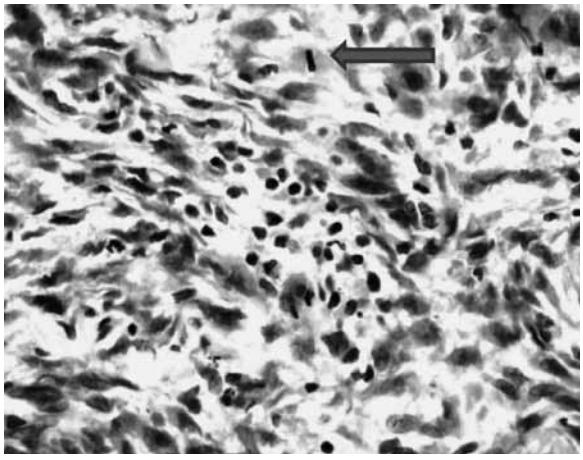
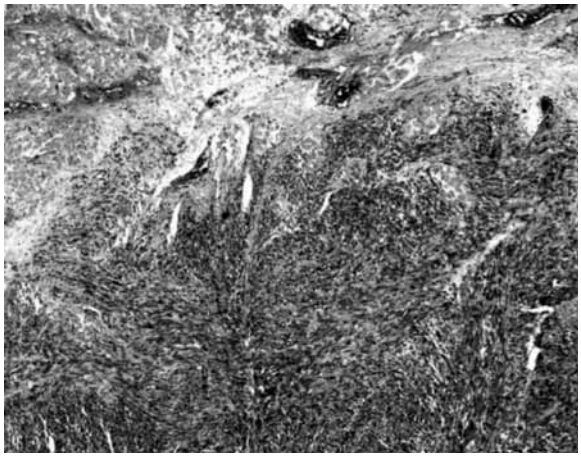


Figure 5 – Infiltrative border (upper field).



DISCUSSION

Orofacial NF is a benign lesion characterized by a progressive or rapid fibroblastic proliferation, possibly in response to local injury or infection[19]. It can affect any age group. Recently, orofacial NF has been found to predominantly affect adults in their 4th to 5th decades of life with no gender or race predilection[1]. Children and young adolescents are less likely to be affected[3,7,9,13,24], although our patient fits into this category.

The clinical presentation and growth rate of orofacial NF are nonspecific. Clinically, it appears as a solitary, mobile, firm, non-encapsulated, well-circumscribed mass with variable growth rates. These lesions are frequently associated with pain due to perineural extension[1,3,14,19,20,25]. Depending on the anatomic location, some lesions maybe ulcerated or affect the adjacent soft and hard tissues[1,11,13,14,26]. Moreover, these lesions attain an average size of 2 cm at the time of diagnosis[1,6]. However, they can reach up to 5 cm in size[4], especially in deeper planes or when they go unnoticed by the patients[1,5,13]. Dayan et al.[1] described two types of growth rapidity in their study: a progressively growing type, and a suddenly rapid growing type. The former is the most common type as reported by the patients, while the latter is less common and can be spontaneous or in response to an insult. Also, they described these lesions as an exophytic, bulging, or deeply located types[1]. In this report, the subcutaneous lesion initially was a progressively growing one. This growth was suddenly accelerated after an insult (i.e. FNA). As a result, it attained a large size over a short period of time (>3cm) in association with pain, bulging into the oral cavity, and extended into the floor of the nasal cavity.

The pathogenesis of NF is uncertain. However, many authors believe it is a self- limiting, reactive lesion that is stimulated by a local injury or infection[1,9,13,21,27-35]. Apparently, this hypothesis is based on two evidences: (1) the presence of these lesions at certain anatomic locations vulnerable to repetitive trauma such as the zygoma, tongue, buccal mucosa and others[3,4,9,13], and (2) the histologic features often mimic atypical reparative granulation tissue[9]. Moreover, many reports previously indicated the presence of these lesions for at least a month before the patients noticed an abnormal growth[1,6]. Others identified the causation prior to the development of these lesions[1,13,36]. In this report, it is clear that trauma was the inciting factor, and thus, we strongly believe that trauma somehow playas a role in the development of NF.

The sum of short history, rarity of the lesion in orofacial region, non-specific clinical presentation, histologic variations, and variability in anatomic locations and epidemiologic data make the diagnosis of orofacial NF very difficult. Unfortunately, many cases of NF were misdiagnosed as more aggressive ones, mainly sarcomatous lesions[1,9,25]. Certainly, careful histologic assessment is the best available diagnostic tool to distinguish this lesion from more aggressive counterparts. However, recent advancements in radiography are promising. Several studies have demonstrated characteristic radiographic findings suggestive of these lesions, based on their anatomic locations and histologic subtypes. Thus, they recommended including orofacial NF in the differential diagnosis list[11,12,16,37]. On one hand, lesions of subcutaneous or submucosal origins with myxomatous or intermediate histologic subtypes appear as well defined homogenous masses on CT scan[5] and pick up higher signal intensity than that of the surrounding tissues on T2 weighted MRI[11,12,16,34,37,38]. On the other hand, intra-muscular

lesions and the ones with fibrous histologic subtype were poorly defined on CT scan[5], picked up markedly low signal intensity on T2 weighted MRI, and frequently showed more radiographic aggressive features[12,16,34,37,38]. Thereafter, Kim et al.[11] suggested that NF should be included in the differential diagnosis list when a superficial mass in the orofacial region shows moderate to marked enhancement on CT scan and MRI images, especially when there is a history of rapidly growing mass in response to trauma[11]. Moreover, Sharma et al.[12] proposed that whenever there is a fascial mass lesion with thickening of the fascial septa, NF should be suspected.

Recently, NF of head and neck region has been demonstrated histologically to be locally or superficially infiltrative into the surrounding skeletal muscles. Weinreb et al.[23] reported that superficial involvement of adjacent skeletal muscles by NF was demonstrated in 30% of their cases. This was attributed to superficiality of skeletal muscles in head and neck region, and due to lack of prominent fascial planes, separating these lesions from the adjacent skeletal muscles. Our case carries similar histologic finding as well. We also think that lack of true capsule may allow this unusual infiltrative behavior of these lesions, especially when attain larger sizes. Consequently, this may explain the difficulty of complete excision of larger lesions in less accessible locations. Consequently, higher recurrence rate maybe expected despite its rarity (<1%)[9].

In conclusion, clinicians must be aware of the fact that the incidence of orofacial NF increased within the past decade. This is in conjunction with slight changes in epidemiologic data, clinical behavior, and radiography.

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