Nodular Fasciitis in the Buccal Mucosa: A Case Report

Tetsuro Ikebe, Yuichi Ogata, Yasuo Takamune, Kazutoshi Ota, Takehisa Obayashi and Masanori Shinohara

Department of Oral and Maxillofacial Surgery, Sensory and Motor Organ Sciences, Faculty of Medical and Pharmaceutical Sciences, Kumamoto University
(Chief: Professor Masanori Shinohara)

Abstract: A case of nodular fasciitis which arose from the buccal submucosal region is reported. One week after an incisional biopsy, the lesion enlarged alarmingly and protruded from the submucosa. Although a sarcoma was suspected because of rapid growth, the diagnosis of the biopsy was nodular fasciitis showing a haphazard arrangement of plump fibroblasts without atypical mitoses. After complete resection, no signs of recurrence were seen.

Key words: nodular fasciitis, oral cavity, immunohistochemistry

Introduction

Nodular fasciitis is a reactive, non-neoplastic lesion in which fibroblast-like cells proliferate rapidly to form a fibrous mass, and is thought to originate from muscular fascia. While the lesion is most common in the upper extremity with the forearm being predisposed, 13% of all cases occur in the head and neck region. The incidence in the oral cavity is extremely rare.

Since nodular fasciitis in the head and neck often develops adjacent to a bony prominence such as the zygomatic arch and the angle of mandible, the reactive growth of fibrous tissue is thought to be initiated by trauma followed by inflammation. Because of its rapid growth and haphazard proliferation of plump spindle-shaped fibroblasts, however, nodular fasciitis is often mistakenly diagnosed as a malignant tumor such as fibrosarcoma and malignant fibrous histiocytoma.

We report a case of nodular fasciitis in the buccal mucosa, which enlarged alarmingly after an incisional biopsy.

Report of a case

A 53-year-old male was referred to our department because of a dull pain in the right cheek during mastication. The symptom had appeared a month earlier. The patient had no history of oral injury. Clinical examination revealed a non-visible, but palpable well-demarcated mass under the right buccal mucosa. The mass was firm and fixed to the underlying subjacent tissues. The overlying mucosa was movable and showed almost normal appearance except for a small erosion facing the right lower second molar (Fig. 1 (A)). There were no signs of odontogenic infection. The skin of the right cheek was intact.

Magnetic resonance images (MRI) demonstrated a well-limited, homogeneous mass in the right buccal submucosal region with high signal intensity in the T1-as well as T2-weighted images (Fig. 2). The lesion seemed to be attached to the anterior border of the masseter muscle.
Because the clinical and image findings suggested a tumorous lesion, an incisional biopsy was performed. The histological diagnosis of the biopsy was granulation tissue or, alternatively, nodular fasciitis. One week later, at the second visit, the lesion had enlarged more than expected and protruded from the buccal mucosa (Fig. 1 B) as if the biopsy had triggered the lesion to grow. The size of the mass was 32 $\times$ 22 mm. The rapid increase in size led us to suspect a sarcoma. A second biopsy was undertaken since we were concerned that the first biopsy failed to target the lesion correctly. The second biopsy was also diagnostic of nodular fasciitis.

We resected the lesion with the surrounding tissues including a part of the buccinator, masseter and periosteum of mandible (Fig. 3), even though simple local excision is the recommended treatment and the prognosis is excellent$^{1,3}$. It was non-encapsulated and fixed to the anterior border of the masseter, suggesting that its origin was the fascia of masseter.

The histology showed a haphazard arrangement of spindle-shaped fibroblast-like cells in a myxoid matrix.
Mitotic figures without atypia were common and abundant vascular vessels and inflammatory cell infiltration were also observed (Fig. 4 (A) (B)).

Immunohistochemical staining indicated that the fibroblast-like cells in nodular fasciitis were positive for α-smooth muscle actin (SMA) and vimentin (Fig. 5 (A) (B)). There were numerous α-SMA-positive small blood vessels in the lesion (Fig. 5 (C)).

After surgery, the mouth opening of the patient was limited because of the buccal scar, but recovered to the normal range a month later. No evidence of recurrence was seen at the one-year follow-up visit.
Nodular fasciitis is a benign lesion of uncertain etiology, and is also known as pseudosarcomatous fasciitis or pseudosarcomatous fibromatosis. The most common localizations are the upper extremity (49%), trunk (18%), head and neck (13%) and lower extremity (17%). Nodular fasciitis has been reported to affect males and females equally, commonly seen in young adults with 85% younger than 50 years of age, and usually has an average diameter of around 2 cm.

On the basis of its anatomic location, nodular fasciitis can be categorized into three types: subcutaneous, intramuscular, and intermuscular types. Histologically, it can also be classified into myxoid, cellular, and fibrous subtypes, and is thought to mature from myxoid to fibrous subtype. The present case seems to be categorized into subcutaneous (subcutaneous), cellular subtype.

Although ultrasound, CT and MRI are commonly used to evaluate nodular fasciitis, there are no reports describing specific image appearances for this lesion. Among these imaging modalities, however, MRI may present more information about it. In the MRI, nodular fasciitis of the upper extremity and trunk appeared homogeneous, hyperintense to muscle on T1-weighted images and homogeneous with signal intensity greater than cutaneous fat on T2-weighted images. The present case was also hyperintense on T1-weighted as well as T2-weighted images, suggesting that the image of nodular fasciitis in the oral cavity is similar to that in the upper extremity and trunk.

As in our case, Davies et al. reported the nodular fasciitis in the parasymphyseal region of the lower jaw increased in size after an incisional biopsy. Although surgical intervention may stimulate it to proliferate, preoperative biopsy seems to be inevitable to make a definitive diagnosis of nodular fasciitis. Because of its rarity in the oral cavity, oral surgeons may suspect the rapidly growing nodular fasciitis of a sarcoma. It is important to carefully diagnose a mass that rapidly grows after biopsy to avoid the unnecessary oversurgery of nodular fasciitis.

There are no specific histopathological markers of nodular fasciitis to aid the diagnosis. Several immunohistochemical studies demonstrated that the spindle fibroblastic cells in nodular fasciitis were positive for SMA and vimentin, but not desmin and cytokeratin, implying that their origin was myofibroblasts. The immunostaining in the present study also showed that the fibroblast-like cells expressed SMA and vimentin. In addition, the SMA-positive vessels were abundant in nodular fasciitis. While the increasing number of blood vessels may feed the fibroblast-like cell and enable to proliferate rapidly, the pathological relationship between spindle fibroblastic cells and vascular smooth muscle cells remains unknown.

References