A Unique Case of Nodular Fasciitis in the Submandibular Gland Mimicking Pleomorphic Adenoma

Itay Chen¹, Daria Kozlova², Geoffrey Pollack¹ and Jean-Yves Sichel¹

¹Department of Otolaryngology, Head and Neck Surgery, Shaare Zedek Medical Center, Jerusalem, Israel
²Pathology Institute, Shaare Zedek Medical Center, Jerusalem, Israel

Abstract

We present a case of a 29-year-old woman with rapid enlargement of her submandibular gland over a 6-month period. After imaging and Fine Needle Aspiration Cytology (FNAC) a probable diagnosis of pleomorphic adenoma was made. Excision was performed. The histopathological report diagnosed the tumor as nodular fasciitis. Nodular fasciitis is a rare entity responsible for 0.0025% of tumors in the head and neck. It is especially rare in the salivary glands and to our knowledge; there has been no report in the literature of nodular fasciitis infiltrating the submandibular gland. In this paper we will discuss the histology and imaging of NF in the submandibular gland and its resemblance to pleomorphic adenoma.

Introduction

Nodular fasciitis is a rare, self-limiting benign tumor consisting of fibroblasts and myofibroblasts. It can occur anywhere in the body with the highest incidence in the extremities followed by the head and neck region with an incidence of 20% [1]. Trauma or infection is thought to be the trigger of the fibroblast proliferation although the exact pathogenesis is still unknown. It usually presents as a painless rapidly growing mass that spreads along the muscular fascia into the subcutaneous tissue and sometimes into the underlying muscle. Due to its rapid growth, its high mitotic activity and pleomorphic picture NF can be easily mistaken for a malignant tumor. Perioperative Fine Needle Aspiration Cytology (FNAC) and imaging such as CT and especially MRI can help in making a presumptive diagnosis [2,3].

Case Report

A 29 year-old generally healthy women were admitted to our department with a 6-month history of a rapidly growing mass in the right submandibular area. The mass was not painful; there was no history of trauma or infection and no history of smoking or alcohol use. The patient did not report fever, weight loss or pain with swallowing. On physical examination a 2 cm, smooth, firm, non-tender mass was palpated at the posterior aspect of the right submandibular triangle with no palpable lymph nodes. Laboratory studies were normal.

Neck ultrasonography revealed a 13 × 20 × 23 mm solid tumor within the right submandibular gland as well as benign appearing cervical lymph nodes. FNAC was then performed. The first attempt was non diagnostic. In the second FNAC specimen (alcohol fixation) blood and small amount of hyalinic material with a few spindle cells were seen. This was read by the cytopathologist as suspicious for pleomorphic adenoma, although a mesenchymal origin could not be ruled out.

Prior to surgery an MRI of the neck with contrast was obtained. This showed a well-demarcated mass measuring 2.4 × 2.0 × 1.1 cm in the right submandibular gland, accompanied by mild enlargement of the gland. The mass was hyper-intense in T2 weighted image, relative to skeletal muscle (Figure 1A) and was hypo-intense on T1 weighted image (Figure 1B). It showed homogeneous enhancement following gadolinium injection (Figure 1C) and hyper-intensity in Apparent Diffusion Coefficient (ADC) map relative to the normal gland. This MRI pattern was consistent with pleomorphic adenoma.

Both the cytology and MRI were in concordance with the probable diagnosis of pleomorphic adenoma. A right submandibulectomy was performed. The surgery was uneventful. The pathology report established the diagnosis of NF within the submandibular gland. The histology showed plump, immature-appearing fibroblasts and myofibroblasts (Figure 2A). The cells were arranged in short, irregular bundles and fascicles with scattered lymphoid cells and extravasated erythrocytes (Figure 2B). The specimen stained positive with alpha actin showing a sub-membranous "tram
Very little is described regarding the MRI image of fibrillar intercellular material may be seen in both cases [5]. In our case, the presence of high ADC value in salivary gland tumors can differentiate benign from malignant tumors [15].

The MRI picture seen in our patient is similar both to the general image of NF in the skeletal system mentioned above and to the characteristics of pleomorphic adenoma on MRI. So like cytology, the MRI pattern of NF is similar to that of Pleomorphic Adenoma (PA).

A previous publication compared MRI and FNAC in differentiating between benign and malignant parotid masses, the combination of MRI and Diffused Weighted Image (DWI) with ADC calculation had similar diagnostic values for determining the specific histological types of common parotid masses as FNAC [16].

**Conclusion**

We describe the first documented case, to our knowledge, of NF within the submandibular gland. Workup prior to surgery included FNAC and MRI. The FNAC was consistent with but not conclusive for the diagnosis of PA since it did not differentiate clear cut between PA and tumor of mesenchymal origin. NF is a self-limited benign process that does not always require surgical removal while PA is a benign tumor treated with surgical extirpation. Differentiating between these...
two pathologies is important since NF can spontaneously regress and surgery can be avoided. Currently there is no non-invasive modality that can provide a definite diagnosis of NF in salivary glands. Open biopsy is not a standard of practice in salivary gland tumor diagnosis due to exacerbation of local spreading and its invasive nature. Core biopsy as a minimal invasive technique can aid us in the final diagnosis and in deciding the necessity of surgery. Thus it is important to exhaust our diagnostic possibilities and also regard the less common etiologies in order to achieve the most favorable outcome for the patients.

Acknowledgments

Department of Radiology, Shaare Zedek Medical Center, Jerusalem, Israel.
Pathology Institute, Shaare Zedek Medical Center, Jerusalem, Israel.

References