American Journal of Otolaryngology and Head and Neck Surgery

Case Report
Published: 25 Apr, 2018

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

Itay Chen[1], Daria Kozlova[2], Geoffrey Pollack[1] and Jean-Yves Sichel[1]

[1]Department of Otolaryngology, Head and Neck Surgery, Shaare Zedek Medical Center, Jerusalem, Israel

[2]Pathology 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

OPEN ACCESS

Correspondence:
Itay Chen, Department of Otolaryngology, Head and Neck Surgery, Shaare Zedek Medical Center, Jerusalem, Israel,
E-mail: chenitay2@gmail.com

Received Date: *31 Mar 2018*
Accepted Date: *21 Apr 2018*
Published Date: *25 Apr 2018*

Citation:
Chen I, Kozlova D, Pollack G, Sichel J-Y. A Unique Case of Nodular Fasciitis in the Submandibular Gland Mimicking Pleomorphic Adenoma. Am J Otolaryngol Head Neck Surg. 2018; 1(1): 1002.

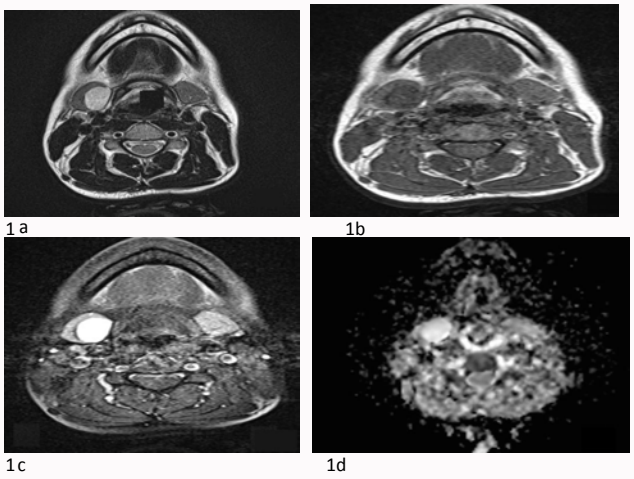

Figure 1: MRI axial section of the neck showing NF in the right submandibular gland with similar features of PA.
A: T2 weighted image showing hyper intensity of the tumor within the right submandibular gland.
B: T1 weighted image showing hypo-intensity of the tumor.
C: A well demarcated hyper-intensity of the tumor Post Gadolinium Injection.
D: ADC map showing hyper-intensity compared with the normal submandibular gland on the left side.

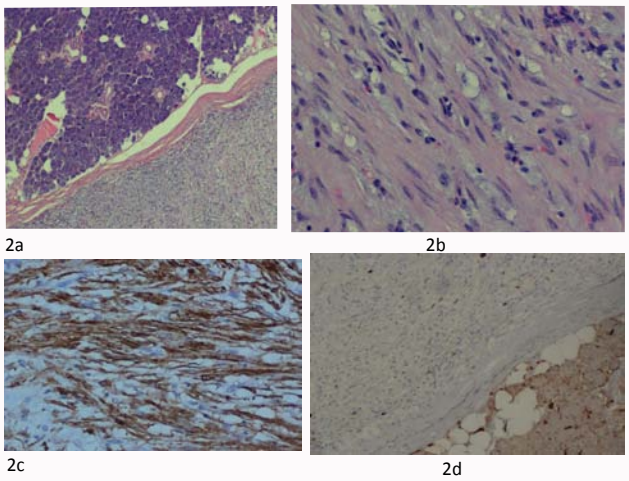

Figure 2: Histologic slides of specimen post right submandibulectomy.
A: 4 X magnification (H&E): left upper field –submandibular gland, right lower field – nodular fasciitis.
B: 20 X magnification (H&E) of the NF tumor showing immature fibroblasts and myofibroblasts.
C: 'Tram track' pattern with alpha actin stain of the NF tumor.
D: S-100 does not stain NF (left upper field) but stains submandibular gland (right lower field).

track" pattern that is characteristic for myofibroblasts (Figure 2C). While the submandibular gland stained with S-100 the lesion did not (Figure 2D).

Discussion

NF is a benign self-limiting tumor which can arise anywhere in the body. The most common areas it is found in are the upper extremity with an incidence of nearly 46%, followed by the head and neck with a 20% incidence and the trunk and lower extremities with an incidence of 18% to 16% [1]. A complete spontaneous resolution of the tumor had been described [4]. NF is very rare within the salivary glands with majority of the cases found in the parotid gland [5-7]. To our knowledge, NF arising from submandibular gland has not been documented before. Although one case report had been published entitled "NF in the submandibular gland" the text revealed that the tumor was located in parotid gland [8].

FNAC is a widely accepted diagnostic tool used in the workup of salivary gland masses differentiating between benign and malignant tumors due to its accuracy and its minimally invasive quality. The diagnostic accuracy of FNAC is reported to be between 86% to 98%, sensitivity of 62% to 97.6% and specificity of 94.3% to 100% [9,10].

Pleomorphic adenoma is the most common major salivary tumor but on FNAC it can be mistaken for a variety of tumors due to its histologic variability [9]. There are many similarities between NF and pleomorphic adenoma on FNAC. Both tumors may demonstrate bipolar spindle cells with central nuclei, loosely dispersed uniform plasmacytoid cells with round/ovoid eccentric nuclei and eosinophilic cytoplasm. Furthermore myxoid background and tufts of fibrillar intercellular material may be seen in both cases [5]. In our case the FNAC favored the diagnosis of pleomorphic adenoma but emphasized that other mesenchymal tumors could not be ruled out.

Although MRI is a well-accepted modality for the diagnosis of salivary gland lesions, very little is described regarding the MRI image of NF in salivary glands in general and within the submandibular gland in particular. MR images of NF in the skeletal system generally appear hyper-intense to skeletal muscle on T2-weighted and iso/hypo-intense on T1-weighted MR images. The vast majority of the lesions are hyper-intense following gadolinium administration [11].

Diffusion-Weighted Imaging (DWI) is an important tool used mostly in the examination of central nervous system pathology. DWI relies on water diffusion characteristics of the tissue, therefore cellularity and cell wall barrier are major factors in the differentiation between different tumors. The impedance of water molecule diffusion can be quantitatively assessed using the Apparent Diffusion Coefficient (ADC) value [12]. The MRI characteristics of pleomorphic adenoma show hypo-intensity in T1 weighted image, hyper-intensity in T2 weighted image, enhancement upon gadolinium injection and relatively high values on ADC map [13,14]. It has been published that 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. Work up 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

1. Goldblum JR, Folpe AL, Weiss SW, Enzinger FM. Enzinger and Weiss's soft tissue tumors. 6th ed. Philadelphia, PA: Saunders/Elsevier; 2014. xiv, 1155 p.
2. Ren SX, Zhang Y. Nodular fasciitis in the orofacial region: a report of three cases. Chin J Dent Res. 2012;15(1):55-9.
3. Borumandi F, Cascarini L, Mallawaarachchi R, Sandison A. The chameleon in the neck: Nodular fasciitis mimicking malignant neck mass of unknown primary. Int J Surg Case Rep. 2012;3(10):501-3.
4. Duncan SF, Athanasian EA, Antonescu CR, Roberts CC. Resolution of Nodular Fasciitis in the Upper Arm. Radiol Case Rep. 2006;1(1):17-20.
5. Saad RS, Takei H, Lipscomb J, Ruiz B. Nodular fasciitis of parotid region: a pitfall in the diagnosis of pleomorphic adenomas on fine-needle aspiration cytology. Diagn Cytopathol. 2005;33(3):191-4.
6. Hwang CS, Lee CH, Kim A, Shin N, Park WY, Park MG, et al. Nodular fasciitis of the parotid gland, masquerading as pleomorphic adenoma. Korean J Pathol. 2014;48(5):366-70.
7. Gibson TC, Bishop JA, Thompson LD. Parotid Gland Nodular Fasciitis: A Clinicopathologic Series of 12 Cases with a Review of 18 Cases from the Literature. Head Neck Pathol. 2015;9(3):334-44.
8. Jang EJ, Park TI, Nam SC, Park JY. Nodular fasciitis in the submandibular gland. Diagn Cytopathol. 2008;36(11):805-8.
9. Zhang S, Bao R, Bagby J, Abreo F. Fine needle aspiration of salivary glands: 5-year experience from a single academic center. Acta Cytol. 2009;53(4):375-82.
10. Rajwanshi A, Gupta K, Gupta N, Shukla R, Srinivasan R, Nijhawan R, et al. Fine-needle aspiration cytology of salivary glands: diagnostic pitfalls--revisited. Diagn Cytopathol. 2006;34(8):580-4.
11. Coyle J, White LM, Dickson B, Ferguson P, Wunder J, Naraghi A. MRI characteristics of nodular fasciitis of the musculoskeletal system. Skeletal Radiol. 2013;42(7):975-82.
12. El Kady RM, Choudhary AK, Tappouni R. Accuracy of apparent diffusion coefficient value measurement on PACS workstation: A comparative analysis. AJR Am J Roentgenol. 2011;196(3):W280-4.
13. Tartaglione T, Botto A, Sciandra M, Gaudino S, Danieli L, Parrilla C, et al. Differential diagnosis of parotid gland tumours: which magnetic resonance findings should be taken in account? Acta Otorhinolaryngol Ital. 2015;35(5):314-20.
14. Habermann CR, Arndt C, Graessner J, Diestel L, Petersen KU, Reitmeier F, et al. Diffusion-weighted echo-planar MR imaging of primary parotid gland tumors: is a prediction of different histologic subtypes possible? AJNR Am J Neuroradiol. 2009;30(3):591-6.
15. Eida S, Sumi M, Sakihama N, Takahashi H, Nakamura T. Apparent diffusion coefficient mapping of salivary gland tumors: prediction of the benignancy and malignancy. AJNR Am J Neuroradiol. 2007;28(1):116-21.
16. Yerli H, Aydin E, Haberal N, Harman A, Kaskati T, Alibek S. Diagnosing common parotid tumours with magnetic resonance imaging including diffusion-weighted imaging vs fine-needle aspiration cytology: a comparative study. Dentomaxillofac Radiol. 2010;39(6):349-55.