CASE REPORT

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Nodular Fasciitis Mimicking a Maxillofacial Cancer

Maksillofasiyal Kanseri Taklit Eden Nodüler Fasiit

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ABSTRACT

Nodular fasciitis is a rare, benign soft tissue mass that is commonly incorrectly diagnosed as a malign lesion due to its rapid and infiltrative growth pattern and histological characteristics. This case report discusses a 52-year-old female patient, who presented with a progressively growing mass of the right mandible within two months, resulting in numbness and tingling around the lips. Possibility of a malignant process was considered based on her complaints and clinical, radiologic, and pathologic findings. A complete excision of the mass that was adherent to the intraoperative marginal mandibular nerve indicated nodular fasciitis. As demonstrated by this case report, nodular fasciitis is a significant entity to consider when faced with similar clinical presentations.

Keywords: Cancer, nodular fasciitis, facial nerve

ÖZ

Nodüler fasiit, hızlı ve infiltratif büyüme paterni göstermesi ve histolojik özellikleri nedeniyle sıklıkla malignite tanısı konulabilen nadir görülen benign bir yumuşak doku kitlesidir. Bu olgu sunumunda, sağ çene kemiğinde iki ay içinde giderek büyüyen kitle yakınması ile başvuran ve dudaklarında uyuşma ve karıncalanma ortaya çıkan 52 yaşında bir kadın hasta tartışılmaktadır. Hastalığın şikayetleri ve klinik, radyolojik ve patolojik bulgularına göre malign bir süreç olduğu düşünüldü. Operasyon esnasında marjinal mandibular sinire yapışık olduğu görülen kitlenin total eksizyonu sonucunda nodüler fasiit tanısı konuldu. Bu olgu sunumunda belirtildiği gibi benzer klinik tablolar ile karşılaşıldığında, nodüler fasiit tanısı dikkate alınmalıdır.

Anahtar Kelimeler: Kanser, nodüler fasiit, fasiyal sinir

Introduction

Nodular fasciitis (NF) is a benign condition that can both clinically and histologically mimic more insidious and sarcomatous lesions. While the pathogenesis of NF has been suggested to be traumatic, infectious, or inflammatory, the role of these factors in the development of lesions is uncertain (1,2). Diagnosis is reported to be mainly between the third and fourth decades, and predominantly in males. In approximately 20% of the cases, NF is localized in the head and neck region. It generally presents as a unilateral painless mass which typically develops within a few weeks in subcutaneous or deeper soft tissue. The diagnosis of NF may be difficult due to various clinical and radiologic findings and troubling histological characteristics (1,3). This article presents an NF case with a rapidly growing lesion of the mandible, resulting in numbness in the lip, and thus being mistaken for a malign lesion based on the clinical examination and also radiological and histopathological findings.

Case Report

A 52 year-old female patient presented with complaints of swelling on the right mandible, which had progressively increased over two months, and numbness and tingling around the lips on the right side. The patient had no history of medication, alcohol or nicotine use, trauma or inflammation in the region, andsystemic disease. In palpation examination a rigid, fixated mass of about 20x10 millimeter (mm) was certainedon the right-side of the mandibular corpus. Facial nerve examination was unremarkable. Subsequent ultrasound has shown a 17x11 mm sized solid mass (suspicious for malignancy) on the anterior of the mandibular bone.

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Consequent contrast-enhanced maxillofacial computerized tomography (CT) revealed a 16x11 mm soft tissue mass that was 4 mm under the skin, with homogeneous contrast and prominent benign lymph nodes in the neck. Fine needle aspiration cytology showed highly cellular smears with a polymorphic appearance (Figure 1). The cells were large and fibroblast-like with well-defined cytoplasmic borders. The nuclei showed pleomorphism with fine, evenly distributed chromatin and prominent nucleoli in many of the cells. A few scattered lymphocytes and neutrophils were present. The background was slightly myxoid. However, excision was recommended for definitive diagnosis. As a preliminary diagnosis of malignancy, fluorodeoxyglucose (18F-FDG) positron emission tomography (PET)-CT was performed (Figure 2). The maximum standardized uptake value (SUV_{max}) of the mass was 12.8. There were benign lymph nodes in the cervical chain. The patient was discussed in multidisciplinary oncology board and they recommended a complete excision of the mass under general anesthesia. During surgery, we noted that the mass located superiorly and very close to the marginal mandibular nerve (Figure 3). The marginal mandibular nerve was preserved as the mass was dissected from the mandible's periosteum and completely removed. No postoperative complication was observed. Histopathology showed benign spindle cells arranged in sheets and fascicles containing moderate-to-abundant fuzzy cytoplasm. Nuclear atypia was absent. Extravasated red blood cells (RBCs) were seen at occasional places with lymphocytic infiltrate. Immunohistochemically, the cellular component displayed reactivity toward vimentin (a fibroblast marker), calponin-b, and smooth-muscle specific actin (Figure 4). The spindle cells of this lesion lack desmin, keratin, CD34, p53, or S100, which aids in securing the diagnosis of this proliferative but benign lesion over the more troubling items on the differential. Based on these findings, it was diagnosed as a NF. The patient remained free of disease at 15 months after surgery. The patient signed an informed consent before the investigation.

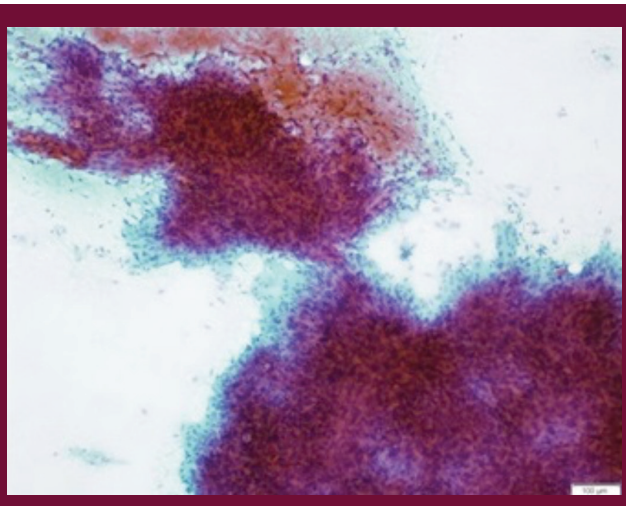

Figure 1. FNAC smear PAP stain 100x (above) cellular smear showing predominantly spindle-shaped cells with low nuclear atypia
FNAC: Fine needle aspiration cytology, PAP: Papanicolaou

Discussion

NF was first characterized and named as pseudosarcomatous fibromatosis by Konwaler et al. (4). Since then, it has had various names such as pseudosarcomatous fasciitis, pseudosarcomatous fibromatosis, proliferative fasciitis, and infiltrative fasciitis. NF was often classified as some form of sarcoma, usually liposarcoma, fibrosarcoma, or rhabdomyosarcoma, before being described as a distinct mass (2).

NF pathogenesis seems to be reactive or inflammatory, involving fibroblastic or myofibroblastic proliferation, rather than truly neoplastic. Some studies suggest that local trauma may induce myofibroblast proliferation. As outlined in case reports of pregnant and lactating women with NF, another assumption is that estrogen receptor stimulation in myofibroblasts may lead to proliferation of those cells (1,5). In our case, the patient had no history of trauma, inflammation, or pregnancy, which suggests that there are some other factors in the pathogenesis of NF that are still unknown.

NF is incorrectly diagnosed due to clinical findings and radiological characteristics similar to malignant lesions.

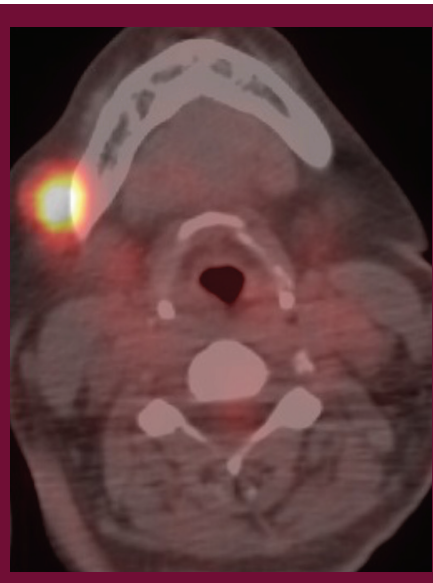

Figure 2. PET/CT with 18F-FDG demonstrates a focal area of uptake by the mandible with an SUV_{max} of 12.8
PET/CT: Positron emission tomography/computed tomography, 18F-FDG: fluorodeoxyglucose

These masses are often misdiagnosed as squamous cell carcinomas, sarcomas, or uncommonly as metastases of other primary tumors. Morbidity of radical surgical procedures can be limited with preoperative diagnosis of this benign tumor (6).

For differential diagnosis, various imaging methods such as CT, magnetic resonance imaging, direct radiography, ultrasound, PET-CT can be performed preoperatively. NF has nonspecific and variable imaging characteristics. In the head and neck, depending on predominant stromal elements, NF may appear as a distinct solid or cystic mass. Lesions may be localized within the subcutaneous space, deeply situated along deep fascia, or embedded within muscle (7). 18F-FDG PET-CT is commonly used for benign/malign differentiation, as well as staging/restaging of various malignances by revealing glucose metabolism (8). However, increased 18F-FDG uptake may also manifest in some benign conditions, including abscess, pulmonary granuloma, tuberculosis, and sarcoidosis. Therefore, false-positive conditions are likely in a clinical setting (9). In our case, high SUV_{max} value led us to believe that the tumor was malignant.

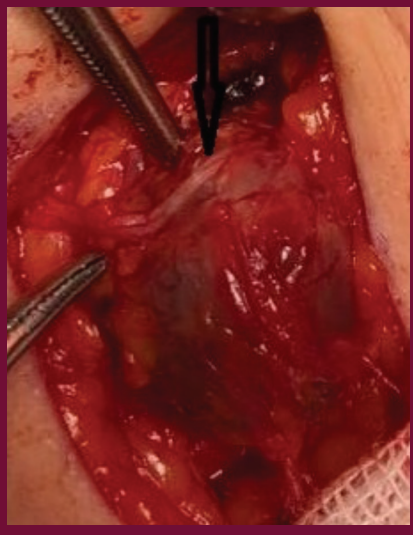

Figure 3. Tumor on the mandible adhering to the marginal mandibular nerve

NF must be clearly distinguished from spindle-cell sarcoma based on the degree of cytologic atypia, prominent in spindle-cell sarcoma. Fibromatosis (desmoid tumor), with dense collagenous stroma and absence of both myxoid areas and extravasated RBCs must also be ruled out (1). Dermatofibroma also shows spindle cell proliferation admixed with epithelioid histiocytes, but also lacks prominent vasculature and RBC extravasation. Proliferative fasciitis is characterized by ill-defined tumor growing along the fibrous septa with large myofibroblasts admixed with immature fibroblast-like spindle cells in a myxoid or collagenous background stroma, but absence of extravasated RBCs. Another differential diagnosis of NF includes benign nerve sheath tumors such as schwannoma, with Antoni A and Antoni B growth patterns, and neurofibroma, with abundant collagen and scant myxoid material, also lacking RBC extravasation (6).

Lenyoun et al. (10) indicated that NF only requires marginal excision without concern of recurrence, and Hseu et al. (3) reported that none of the patients who underwent complete resection developed recurrence. In our patient, who also underwent complete excision, the tumor was adherent to the marginal mandibular nerve and nerve was expanded. On the basis of these outcomes, we strongly recommend complete

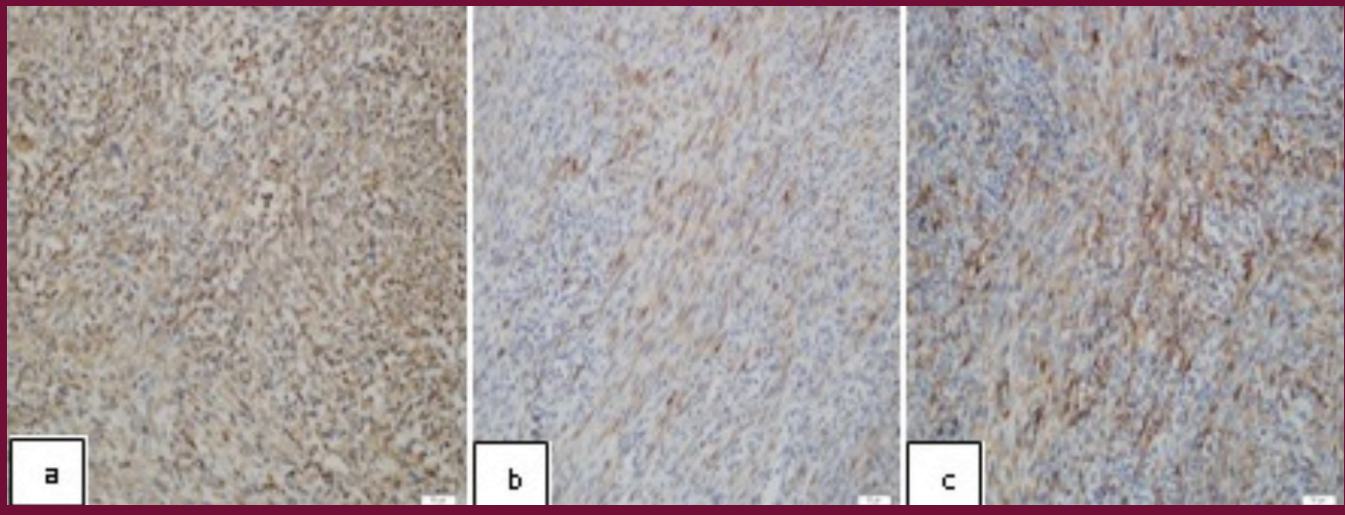

Figure 4. Wide spread positivity for vimentin (a), positive staining of fibroblasts, myofibroblasts for calponin-B (b), and smooth muscleactin (c) stains in sub-membranous "tram-track" pattern characteristic of myofibroblasts

local excision. Furthermore, localization of the mass must be considered because an imprecise procedure may cause an unwanted outcome such as facial paralysis. The patient had no sign of recurrence at 15 months after surgery.

Conclusion

In regard to the clinical and histopathological similarities of the NF, it can be easily misdiagnosed as a malignant tumor. NF should be in mind in the differential diagnosis of the masses in the head and neck area. Aggressive treatment should be avoided in patients who are considered to be diagnosed with NF.

Ethics

Informed Consent: The patient signed an informed consent before the investigation.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: S.B., N.K.T., B.E.E., İ.E.Ç., Concept: S.B., N.K.T., B.E.E., İ.E.Ç., Design: S.B., N.K.T., B.E.E., İ.E.Ç., Data Collection or Processing: S.B., N.K.T., B.E.E., İ.E.Ç., Analysis or Interpretation: S.B., N.K.T., B.E.E., İ.E.Ç., Literature Search: S.B., N.K.T., B.E.E., İ.E.Ç., Writing: S.B., N.K.T., B.E.E., İ.E.Ç.

Conflict of Interest: No conflict of interest was declared by the authors.

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References

1. Dayan D, Nasrallah V, Vered M. Clinicopathologic correlations of myofibroblastic tumors of the oral cavity: 1. Nodular fasciitis. J Oral Pathol Med. 2005;34:426-435. [Crossref]
2. Nair P, Barrett AW, Theodossy T. Oral nodular fasciitis: case report. Br J Oral Maxillofac Surg. 2004;42:360-362. [Crossref]
3. Hseu A, Watters K, Perez-Atayde A, Silvera VM, Rahbar R. Pediatric nodular fasciitis in the head and neck: evaluation and management. JAMAOtolaryngol Head Neck Surg. 2015;141:54-59. [Crossref]
4. Konwaler BE , Keasbery L, Kaplan L. Subcutaneous pseudosarcomatous fibromatosis (fasciitis). Am J Clin Pathol. 1955;25:241-252. [Crossref]
5. Chi AC, Dunlap WS, Richardson MS, Neville BW. Intravascular fasciitis: report of an intraoral case and review of the literature. Head Neck Pathol. 2012;6:140-145. [Crossref]
6. Ren SX, Zhang Y. Nodular fasciitis in the orofacial region: a report of 3 cases. Chin J Dent Res. 2012;15:55-59. [Pubmed]
7. Kim ST, Kim HJ, Park SW, Baek CH, Byun HS, Kim YM. Nodular fasciitis in the head and neck: CT and MR imaging findings. AJNR Am J Neuroradiol. 2005;26:2617-2623. [Pubmed]
8. Asagi A, Ohta K, Nasu J, Tanada M, Nadano S, Nishimura R, et al. Utility of contrast-enhanced FDG-PET/CT in the clinical management of pancreatic cancer: Impact on diagnosis, staging, evaluation of treatment response and detection of recurrence. Pancreas. 2013;42:11-19. [Crossref]
9. Seo M, Kim M, Kim ES, Sim H, Jun S, Park SH. Diagnostic clue of nodular fasciitis mimicking metastasis in papillary thyroid cancer, mismatching findings on 18F-FDG PET/CT and 123I whole body scan: A case report. Oncol Lett. 2017;14:1167-1171. [Crossref]
10. Lenyoun EH, Wu JK, Ebert B, Lieberman B. Rapidly growing nodular fasciitis in the cheek of an infant: case report of a rare presentation. Eplasty. 2008;8:e30. [Pubmed]