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Case Report

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Elastofibroma Dorsi: Two Cases of a Rare Benign Tumor

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Abstract

Elastofibroma dorsi is an uncommon benign and a slowly growing fibroblastic pseudotumor that is typically located at the lower pole in the subscapular region of middle-aged or older individuals. Pathogenesis is still unclear and a matter of debate. The morphology and clinical behaviour indicate that elastofibroma dorsi is a reactive fibromatosis rather than a true neoplasm. Elastofibroma dorsi is mostly described as case reports or case series in the literature due to its rare occurrence. Magnetic resonance imaging can be used as a first-line investigation of the lesion and reveals a lenticular soft-tissue mass with a signal intensity similar to that of skeletal muscle interlaced with strands of fat. We present two cases of elastofibroma dorsi presenting with distinct presentations comprising only back pain revealing a completely normal laboratory profile. Both cases revealed mild to moderate uptake on FDG PET-CT while the final diagnosis was achieved by CT and MR imaging. Our goal is to identify the diagnostic challenges that arise with this extremely rare benign tumor, as well as to draw a roadmap for presenting unique treatment options for each patient.

Keywords:
Elastofibroma dorsi, Mesenchymal tumours, MRI, PET/CT

Introduction

Elastofibroma dorsi is a rare, slow growing, ill-defined soft tissue benign tumour of the chest wall that is thought to emerge due to chronic mechanical friction between the scapula and the thoracic wall, leading to fibroelastic tissue proliferation that emerges more commonly in individuals with involved physical labor or repetitive shoulder movements. It is commonly located beneath the rhomboid major and latissimus dorsi muscles [1-6]. Elastofibroma dorsi is usually located in the infrascapular regions, deep to the serratus anterior and latissimus dorsi muscles. Unilateral masses have a slight right-sided predilection while up to one third of these tumors are bilateral. The lesions are seen in patients over the age of 50 years and not uncommonly mistaken as a malignant tumor because of their size and location deep to the periscapular muscles. Recognition of the lesion is crucial because the differential diagnosis includes malignant tumors [7-10]. We report two cases of bilateral elastofibroma dorsi where CT and MR imaging appeared as the hallmark of final diagnosis while PET/CT permitted the diagnosis of this rare, benign tumor, that may have eliminated preoperative histological examination. Due to its rare occurrence, elastofibroma dorsi emerges as a diagnostic and therapeutic challenge in clinical practice.

Case I

A 70-year-old male dentist presented with two painful masses located bilaterally in the right and left inferior periscapular region. The masses had enlarged slowly over the previous twelve months. The pain progressively increased in intensity and radiated back bilaterally. Personal and family history did not reveal any disease of concern. The patient was a non-smoker and did not use any drugs or alcohol. Physical examination revealed a tender, firm mass with a diameter of 70 mm in the left and a tender, firm mass with a diameter of 50 mm in the right infrascapular region. Initial investigations showed a normal blood count, bone profile, inflammatory markers, and a normal chest radiograph. MRI of the thorax revealed two well-defined soft tissue lesions, with a striated appearance, measuring 70 mm×20 mm on the left and 50 mmx15 mm on the right inferior scapular region (Figure 1). A needle aspiration biopsy was performed on both sides. The smear was characterized by a mixture of uniform spindle cells and very few mature adipocytes, with fragments of collagen bundles, and fibers. PET/CT images showed poorly circumscribed, bilateral soft tissue masses between the inferior tips of the scapulae and chest wall, with low-grade, diffuse ^{18}F-fluorodeoxyglucose uptake (Figure 2). Bilateral surgical excision of the lesions was performed. Postoperative histopathological examination of the resected tumors revealed scant fibroelastic proliferation, with abundant hyalinized collagen, and entrapped mature adipose tissue, consistent with the diagnosis of elastofibroma dorsi. The patient was asymptomatic after surgery without any recurrence during follow-up.

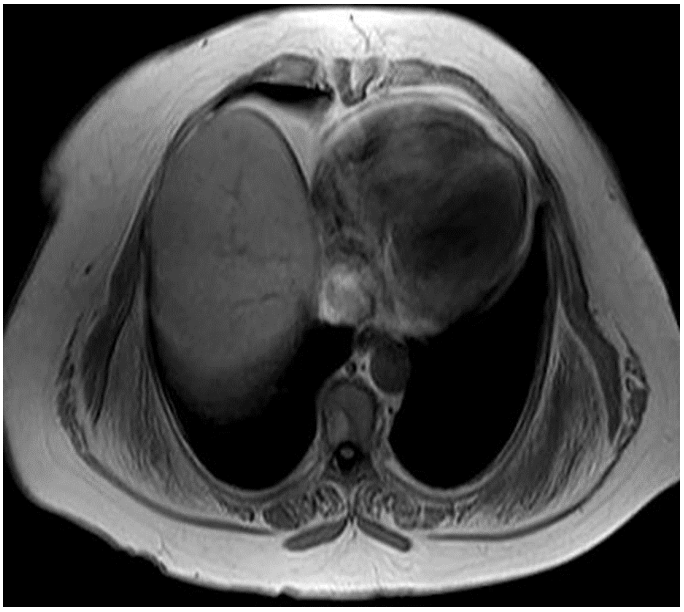

Figure 1: MRI revealing bilateral showing alternating fibrous and fatty components.

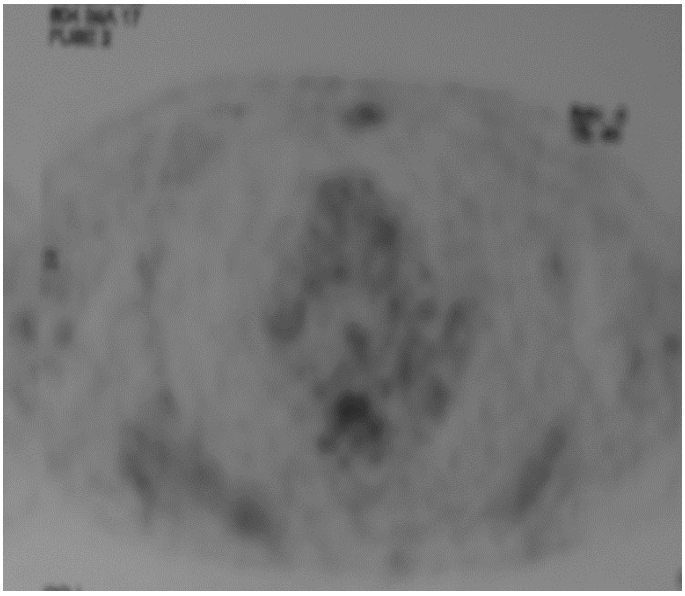

Figure 2: PET/CT revealing low, diffuse 18F-fluorodeoxyglucose uptake on both sides in the infrascapular and the subscapular region.

Case II

A 59-year-old woman presented with a six-month history of intermittent but progressive discomfort in the right scapular region exacerbated by arm movements. She denied any trauma or systemic symptoms. She had a smoking history of 30 p-year. The patient had a total thyroidectomy 20 years ago and had commenced on 100 mg/day levothyroxine sodium treatment. Family history revealed type II diabetes mellitus in the mother and lung carcinoma in the father. Physical examination displayed a firm, non-tender mass deep to the inferior angle of the right scapula, palpable during shoulder abduction. Complete blood count and serum biochemistry were within normal limits except for a slightly elevated CRP level of 15.6 mg/L. Chest x-ray was normal chest while thorax CT revealed two well-defined soft tissue lesions, with a striated appearance, measuring 70 mm×20 mm on the right inferior scapular region (Figure 3). Magnetic resonance imaging showed a well-defined, heterogeneous mass on both sides with characteristic alternating fatty and fibrous bands suggestive of elastofibroma dorsi without any evidence of invasion into adjacent structures. PET/CT revealed low grade, diffuse ^{18}F-fluorodeoxyglucose uptake in this location. Given her mild symptoms and the benign nature of the lesion, a conservative approach was taken. The patient was counseled regarding the condition, and physical therapy was recommended to alleviate discomfort. At a six-month follow-up, her symptoms had improved without further intervention.

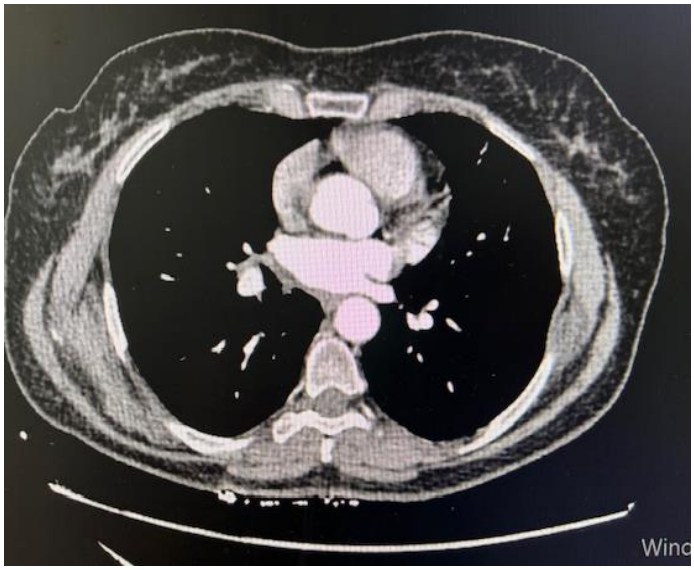

Figure 3: Thorax CT showing soft-tissue masses located in the infrascapular and subscapular region with attenuation similar to the adjacent skeletal muscle.

Discussion

Elastofibroma is a benign, slow-growing mesenchymal soft tissue pseudotumor first described in 1961 by Jarvi [11]. The tumor predominantly affects the elderly individuals and is more common in women. It arises from repetitive mechanical friction between the scapula and thoracic wall that induces fibroelastic proliferation [1-3]. Although often asymptomatic, patients may present with localized discomfort or a sense of fullness in the back. Bilateral involvement occurs in up to 66% of cases [2,12,13]. An incidental prevalence of 2% was found in an elderly population examined using chest CT while an autopsy series found a frequency of 11.2% in men and 24.4% in women [14,15]. The characteristic location is between the chest wall and the inferior tip of the scapula. Bilateral involvement occurs in only 10% of patients [5]. Most patients are asymptomatic and present with painless swelling. Less than 10% of patients present with back pain in the scapular area [12,13]. Plain radiographs may be normal or may show soft tissue density in the periscapular region. Ultrasound demonstrates a well-defined multi-layered pattern of hypoechoic linear areas of fat deposition intermixed with echogenic fibroelastic tissue that may be a useful non-invasive imaging modality [16,17]. On CT, elastofibroma appears as a poorly circumscribed soft-tissue mass with ill-defined margins and an attenuation like that of muscle. The mass appears as alternating fibrous and fatty components. Strands of lower density attributed to fat can be seen within the lesion [14-17]. Although the borders of these masses are relatively well defined, no capsule can be identified in MR. The lesions show relatively low signal intensity similar to muscle on T1- and T2-weighted images. Interlaced fat is seen

as strands of high signal intensity within these hypointense lesions [18,19]. PET/CT image reveals a region of bilateral and asymmetric FDG accumulation that reveals mild or moderate metabolic activity on PET/CT imaging (20-22). The differential diagnosis includes desmoid tumors, neurofibroma and liposarcoma.

Elastofibroma is a rare, slow growing and benign soft tissue tumour of the chest wall. It is commonly located beneath the rhomboid major and latissimus dorsi muscles. Exact prevalence is unknown as most are detected incidentally. Elastofibroma is thought to be associated with long-standing microtrauma that is more commonly observed in elderly women, laborers, dentists, or other manual workers. Pathologically, the tumor shows proliferation of fibroelastic tissue with excessive abnormal degenerated elastic muscle fibers and appears as a well circumscribed lesion, comprised of mature fat, fibrous tissue that comprises characteristic abnormal eosinophilic looking elastic fibers. Elastotic degeneration of collagen or abnormal elastotic fibrogenesis is the hallmark pathogenetic sequela of elastofibroma and active neovascularization or endothelial mesenchymal transition also plays a potential evolutionary role [23]. Most recently, chromosomal abnormalities have been described in a few cases, including those for chromosome 1, suggesting a neoplastic nature for this tumor [24]. Biopsy should, therefore, be undertaken as a confirmatory diagnostic procedure to exclude sarcoma or other soft tissue malignant tumors. The differential diagnosis comprises other malignant, metastatic tumors or even collagen vascular and autoimmune disorders.

We present two cases of elastofibroma dorsi with an unique symptom of back pain displaying an atypical clinical profile that has led to a diagnostic challenge. Identification of elastofibroma poses great difficulties, because specific symptoms, definitive physical examination findings, or imaging manifestations are usually lacking and if they are present they pose an extremely non-specific nature simulating many other diseases. The only symptom exclusive for elastofibroma was noted to be the back pain, although a hallmark symptom, is one of the most frequent complaints, especially in the elderly patients in clinical practice. Back pain, the hallmark of elastofibroma, comprises many different etiologic disorders that constitutes an extremity for an inexorable differential diagnostic assessment, and emerges as the greatest obstacle to a definitive diagnosis. Imaging, particularly the MR, is critical for diagnosis, revealing the pathognomonic pattern of alternating fibrous and fatty tissue [16-19]. PET/CT, on the other hand, displays minimal or moderate ^{18}FDG uptake that does not provide a specific and a precise designation. Biopsy is required to confirm a definitive final diagnosis in most cases. Symptomatic lesions or those that present with diagnostic uncertainty warrant surgical excision which also introduces a simultaneous curative option for the patients. Radiotherapy can be used in high-risk patients for surgery. Asymptomatic lesions can be managed conservatively with regular monitoring.

Conclusions

Elastofibroma dorsi is a rare benign tumor with a predilection for the subscapular region of middle aged or elderly individuals. Pathogenesis is still unclear and a matter of debate. Thorax CT may be useful as the initial diagnostic step. MR imaging appears to be the basic imaging modality for elastofibroma diagnosis which reveals a lenticular soft tissue mass with a signal intensity of skeletal muscle interlaced with strands of fat. PET/CT is usually non-diagnostic displaying minimal or moderate FDG uptake that may discriminate a malignant process. Biopsy is required if pathognomonic criteria are equivocal. Awareness of the clinical profile and the imaging features is essential for an accurate diagnosis of elastofibroma dorsi. Treatment comprises a conservative approach while wait and see attitude constitutes a reasonable convergence. These two cases underscore the consequence of recognizing the distinctive and exclusive imaging features of elastofibroma dorsi to avoid unnecessary interventions and ensure appropriate management. Conservative management is often sufficient for asymptomatic patients while surgery is reserved for symptomatic or cases that carry a diagnostic uncertainty.

Author contributions:

Cuneyt Tetikkurt contemplated and wrote the case report.
Muammer Bilir prepared the laboratory findings of the patient.
Nihal Bayar presented the imaging manifestations of the cases.
Umit Seza Tetikkurt wrote the pathological mechanisms involved for elastofibroma dorsi development.
All authors declare that they do not have any conflicts of interest associated with the case report. Authors confirm that there does not exist any supporting or funding agencies for this case study.

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