

Elastofibroma dorsi: an unusual cause of shoulder pain

Elastofibroma dorsi: Nadir görülen bir omuz ağrısı nedeni

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Amaç: Elastofibroma dorsi (ED) nedeniyle cerrahi tedavi uygulanan hastalar değerlendirildi.

Çalışma planı: Çalışmaya ED tanısı konan ve yakınmaların sürmesi nedeniyle cerrahi tedavi uygulanan 13 hasta (11 kadın, 2 erkek; ort. yaş 54; dağılım 43-74) alındı. Altı hastada sağ, beş hastada sol ve iki hastada iki taraflı tutulum vardı. Tüm hastalarda, omzun öne fleksiyon ve adduksiyon hareketleri sırasında kitle skapula alt köşesinde görünür hale gelmekteydi. Dokuz hastada sırtta şişlik ve ağrı, dördünde ise omuz hareketleri ile oluşan atlama hissi ve omuz ağrısı yakınması vardı. Tanı 10 hastada manyetik rezonans görüntüleme, üç hastada ise bilgisayarlı tomografi ile kondu; hicbir hastaya tanı amaçlı biyopsi uygulanmadı. Cerrahi tedavi olarak, tüm hastalarda marjinal sınırlarda tümör eksizyonu yapıldı. Hastalar, ameliyat sonrası dönemde nüks açısından ultrasonografi ile değerlendirildi. Ortalama takip süresi 32 ay (dağılım 8-90 ay) idi.

Sonuçlar: Tümörlerin hepsi, skapula alt köşesinde, serratus anterior, romboid ve latissimus dorsi kasları arasında, toraks duvarına yapışık bulundu. Çıkarılan kitlelerin ortalama büyüklüğü 9x6x3 cm (dağılım 5x3x1-14x8x3cm) idi. Tanı bütün hastalarda histopatolojik olarak doğrulandı. Tüm hastalarda ana yakınmalar geriledi. Cerrahi sonrasında dört hastada hematom görüldü. Bu hastalar cerrahi girişime gerek kalmadan iyileşti. Hiçbir hastada nüks görülmedi.

Çıkarımlar: Elastofibroma dorsi, klinik olarak az rastlandığı için omuzla ilgili yakınmalarda tanı olarak düşünülmelidir. Yakınmaları olan hastalarda, tümörü marjinal sınırlarda çıkarmak yeterlidir.

Anahtar sözcükler: Fibrom/cerrahi; manyetik rezonans görüntüleme; skapula; omuz ağrısı/etyoloji; yumuşak doku neoplazileri/ cerrahi; göğüs neoplazileri/cerrahi; bilgisayarlı tomografi. **Objectives:** We evaluated patients who underwent surgical treatment for elastofibroma dorsi (ED).

Methods: The study included 13 patients (11 women, 2 men; mean age 54 years; range 43 to 74 years) who were treated surgically for ED that caused persistent symptoms. Involvement was on the right in six patients, on the left in five patients, and bilateral in two patients. All the patients presented with a mass lesion that became apparent at the lower corner of the scapula on shoulder flexion and adduction. The complaints were swelling and pain on the back in nine patients, and a snapping sound on shoulder movements together with pain in four patients. Diagnosis of ED was made by magnetic resonance imaging (n=10) and computed tomography (n=3), with no utilization of preoperative biopsy. Marginal tumor excision was performed in all cases. Evaluation for recurrence was made by ultrasonography. The mean follow-up period was 32 months (range 8 to 90 months).

Results: All the masses were located at the inferior corner of the scapula, with adherence to the thorax between the serratus anterior, rhomboid, and latissimus dorsi muscles. The mean size of the surgical specimens was 9x6x3 cm (range 5x3x1 to 14x8x3 cm). Clinical diagnosis was confirmed by histopathologic examination in all cases. All major complaints resolved after surgery. Hematoma occurred in four cases postoperatively, but resolved without the need for surgical intervention. No recurrence was observed.

Conclusion: Even though ED is a rare clinic entity, it should be recalled while evaluating shoulder pathologies. Marginal excision is adequate for the treatment of patients with sustaining complaints.

Key words: Fibroma/surgery; magnetic resonance imaging; scapula; shoulder pain/etiology; soft tissue neoplasms/surgery; thoracic neoplasms/surgery; tomography, X-ray computed.

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Elastofibroma dorsi (ED) is an uncommon, fibrous tissue oriented soft tissue tumor with slow infiltrative growth, which is non-neoplastic, on the other hand.^[1-5] It was first defined by Jarvi in 1961.^[6] It is generally located at the inferior corner of the scapula, adherent to the thoracic wall between the latissimus dorsi and serratus anterior muscles. ^[2,3,7,8] The autopsy incidence rate has been reported to range from 13% to 17%.^[9] Although it generally occurs unilaterally in women over the age of 50, bilateral involvement may also be observed by 10%.^[10] Its etiology has not been defined yet.^[2] In some studies, it has been stated that ED might occur as a result of the mechanical friction between the chest wall and scapula, as a result of repetitive traumas.^[1,6] It may not cause clinical complaints in almost half the number of patients. It generally causes such complaints as swelling, pain on back and shoulder, and snapping sound on back.^[5,7,11] Painful swellings bigger in size than five centimeters may raise suspicion for the presence of malignity in both the patient and the surgeon.^[2,3] Although it has been stated by some authors that radiological assessment is sufficient for the diagnosis, authors -recommending biopsy in terms of excluding malignity- also exist. [2,3,8,12-15]

There are few clinical studies regarding the diagnosis and treatment of Elastofibroma Dorsi, and still there are discussions on ED. In this study, we have evaluated patients with clinical complaints of ED on whom surgical treatment was applied.

Patients and method

13 patients (11 women, 2 men; mean age 54 years; range 43 to 74 years) whom were treated surgically for ED between the years 2001-2007 have been retrospectively examined. Involvement was on the right in six patients, on the left in five, and bilateral in two patients. The patients' symptoms in the order of occurrence were mass on the back, being unable to sleep due to the burning pain on the back, pain around the shoulder and a snapping sound during shoulder movement. It has been observed that nine patients had back and shoulder pain around four months (range; 1 to 9 months) later, following the occurrence of the mass on the back. Four patients had the complaints of snapping and pain in shoulder movements. In all the patients, the mass became apparent in the lower corner of the scapula during shoulder flexion and adduction.(Figure 1). Diagnosis of ED was made by magnetic resonance imaging (MRI) and computed tomography (CT), with no utilization of preoperative biopsy. Evaluation for recurrence was made by ultrasonography (USG). The mean follow-up period was 32 months (range 8 to 90 months).

During the preparation for the surgery, the patients were positioned prone, in a way to let the affected side's upper extremity move. In the lower corner of the scapula, a transverse surgical incision was made parallel to the ribs. Marginal tumor excision was performed in all cases. During the closure of the surgical wound, the surrounding muscle tissues were fixed to the thoracic wall in an inward direction following the application of an aspirative drain in order to avoid a dead space.

Results

Radiologically in all cases, a mass with irregular margins was observed in the lower corner of the scapula, near the thoracic wall (Figure 2).



Figure 1. Elastofibroma dorsi in the lower corner of the scapula



Figure 2. (a) T₁-weighted MR Image of Elastofibroma Dorsi in a bilateral involvement case. (b) Mass lesion coherent with ED near the thoracic wall in a unilateral case.

All tumors were observed to be surrounded by serratus anterior, rhomboid, and latissimus dorsi muscles in the lower corner of the scapula, adherent to the periosteum of the seventh ribs. Tumor margin was not totally distinguishable in none of the cases, capsular structure wasn't observed. The mean amount of postoperative drainage was 147 ml (range 70 to 300 ml). The mean size of the masses taken out was 9x6x3 cm (range 5x3x1 to 14x8x3 cm) (Figure 3). Parallel to the clinical and radiological results, pathology results of all the patients were found to be coherent with elastofibroma. In nine patients with preoperative back and shoulder pain, the complaints were relieved postoperatively. Four patients with snapping on the back and shoulder pain still had slight pain on the back - although the snapping was relieved.



Figure 3. The excised mass

During the follow-up period, hematoma was observed in four of the patients. In three of them, hematoma resolved itself during the clinical follow-ups. One of the patients was applied hematoma aspiration with an injector once and additional intervention wasn't needed later on. None of the patients had surgicalwound infections and no recurrence developed.

Discussion

Elastofibroma is a benign soft tissue mass the etiology of which hasn't been defined yet.^[2] In some studies, it has been defined as the reactive growth of the fibroelastic tissue as a result of the recurrent traumas between the thoracic wall and the scapula.^[4] It has been emphasised that the progress of ED might be caused by traumas, as it commonly occurs on the more frequently used right side and on heavy workers.^[8] As a result of the examination of 170 patients, Nagamine and Nohara [16] observed family history in 32% of the cases, and they stated that there might be genetic susceptibility. Some authors state that circulatory insufficieny caused by degeneration in the fibrous tissue is the main cause of ED.^[17] Although clinically rare, it has been shown in the autopsy cases over the age of 55 that masses of 3 cm or smaller in size existed in 24.4% of women, and 11.2% of men.^[17] While the men/women rate has been stated to be 2/5; Chandrasekar et al.^[3] stated that 80% of the total cases were men. Bilateral involvement has been observed to be 10%.^[10,18] In our studies, 84.6% of the paients was women, most of whom were housewives, and family history was observed in none of the cases. There was equal distribution in terms of the side of the lesion. These findings show that etiologically, factors other than trauma were also effective.

The typical location of elastofibroma is the lower corner of the scapula. They can also occur in such atypical locations as axilla, mediastinum, stomach. inguinal area, and foot.^[16] In all our cases, the masses were in the typical location. There is no clinical problem in half of the Elastofibroma Dorsi patients.^[5,7,11] As the masses grow in time; the patients start to feel back and shoulder pain, snapping feeling in shoulder motions, and sweeling on their backs while lying. The pain in the shoulder may cause different treatments and diagnoses. Majo et al.^[5] stated that three out of ten patients they had examined had been performed failed treatments previously with the diagnosis of impingement syndrome; and six patients were applied preoperative physiotherapy and steroid injections because of the snapping on the back; and that their complaints were relieved only after the surgical excision of the masses. When the scapula moves upon the mass during the scapulothoracic motions, painful crepitus may occur. This crepitus is strenghtened by the thoracic cavity, and it becomes audible.^[19] Osteochondroma below the scapula, malunion of ribs or scapula, thinning of tissues between scapula and ribs as a result of a muscle atrophy caused by nerve injuries, and the inflammation of the bursal structures around the scapula can be said to be amongst the other etiological reasons of the situation.^[20] In four of our cases, surgical treatment was performed as the complaints prevailed despite the conservative treatment.

There are various evaluations to diagnose Elastofibroma Dorsi. The presence of typical location and bilateral involvement in a woman patient of an old age may raise the likelihood of ED as a referral diagnosis. The mass becomes apparent with the flexion and adduction of the shoulder during the physical treatment. The crucial radiological evaluation method is MRI.^[8,12] The MR Image of a heterogeneous mass with irregular margins is characteristic. In the MRI examination, Malghem et al. ^[12] stated that fibrous tissues in the mass had similar signal intensities to the surrounding muscle tissues, while adipose tissue showed higher signal intensity than the mass; and they reported that these findings were pathognomonic for the mass. Navlor et al.^[8] reported that radiological evaluation was sufficient for the diagnosis. Massengill et al.^[13] stated that biopsy was not necessary and that the right diagnosis could be made through typical clinical and radiological findings. The MRI findings of the patients we studied upon showed similar characteristics to the literature. Solivetti et al.^[21] reported that Ultrasonography was both a sufficient and a cheap method for the diagnosis. In our cases, we used Ultrasonogrophy for recurrence examination in final clinical examinations. Kransdorf et al.^[22] MRI and CT evaluations were coherent with histopathological evaluation.

Lipoma, fibroma, aggressive fibromatosis, desmoid tumor and sarcoma can be considered clinically in the distinguishing diagnosis of Elastofibroma Dorsi. ^[4,14] Local recurrence has not been reported. Generally, malignity can be considered if there is a deeply located mass bigger than 5 cm in size.^[2,3] Alberghini et al.^[23] reported that they observed a high grade spindle-cell carcinoma together with ED in one case and that there might be a connection between the two of them. There are some authors suggesting diagnostic biopsy. Haves et al.^[15] suggest diagnostic core needle biopsy, while Daigeler et al.^[4] suggest open biopsy considering the heterogeneous structure of the mass. Domanski et al. reported that the results of fine needle aspiration biopsy they applied on patients were highly accurate as in diagnostic means, and that surgical biopsy was not necessary. As we thought that clinical examination and radiological evaluation were sufficient, we performed diagnostic biopsy in none of the patients. In the histopathological examination of elastofibroma dorsi, we observed the presence of clear eosinophilic elastic fiber sections - which were small, round-shaped and had regular margins - in the fibroadipose tissue with a myxoid degeneration.

It has been reported by many authors that marginal tumor excision would be sufficient for the treatment of Elastofibroma Dorsi in patients having complaints. ^[1,2,3,5,7,10] There are some authors suggesting diagnostic biopsy for the patients with no complaints.^[2] However, it has been emphasized that biopsy is not necessary for especially old patients whom were diagnosed with bilateral involvement radiologically. Muramatsu et al.^[2] suggested the excision of the masses bigger than 5 cm in size which do not cause any complaints for the patient in case of malignity; and suggested biopsy to confirm histological diagnosis in patients whom were planned to be followed-up. The most frequently encountered post-surgery problems are hematoma and seroma.^[2,4] Performing bleeding control and resting the arm have been recommended during the excision of the mass adherent in a wide area.^[4] In the patients in our studies, the closure of the wide dead space, which occurred following the surgical excision of the mass, was ensured through the fixation of the surrounding muscle tissues to the periosteum of the ribs. Seroma occurred in none of the patients, and the insufficient extremity rest of the patients was considered to be the cause for the issue. In the clinical follow-ups of these patients, hematomas were observed to resolve themselves without any surgical intervention.

As a result, ED is a benign tumor occurring in the lower corner of the scapula in middle-aged patients. As it is clinically rare, it should be considered for diagnosis in complaints regarding the shoulder. Clinical and radiological evaluations are sufficient for diagnosis. Marginal tumor excision shall be adequate for the patients having complaints.

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