

Does elastofibroma dorsi occur more frequently on the same side with the dominant hand?

Baskın kullanılan el tarafında elastofibroma dorsi daha fazla mı gelişiyor?

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ABSTRACT

Background: In this study, we aimed to investigate the correlation between the side of elastofibroma dorsi formation and the dominant-hand of the patients we operated.

Methods: A total of 84 patients with elastofibroma dorsi (12 males, 72 females; mean age: 55.5±7.0 years; range, 43 to 74 years) were retrospectively analyzed between April 2007 and May 2019. Dominant hand, location, size, sex, age, occupations and follow-up of the mass were recorded and the pre- and postoperative Visual Analog Scale scores were noted.

Results: The mean symptom duration was 9.5±7.8 (range, 3 to 48) months. The lesions were located on the right in 49 (58.3%), on the left in 16 (19%) and bilateral in 19 (22.6%). There was no significant difference between the mass size and age, symptom duration and complication development (p=0.129). A significant relation was found between the formation of the mass on the side of the dominant hand (p=0.010). A significant difference was found between the pre- and postoperative Day 90 Visual Analog Scale scores (p<0.001), indicating a significant decrease in the pain scores.

Conclusion: Elastofibroma dorsi has good clinical results after surgical resection. In our series, the patients had significantly more elastofibroma dorsi on their dominant-hand side.

Keywords: Chest wall tumors, elastofibroma dorsi, soft tissue tumors.

ÖZ

Amaç: Bu çalışmada elastofibroma dorsi gelişimi ve ameliyat edilen hastaların baskın eli arasındaki ilişki araştırıldı.

Çalışma planı: Nisan 2007 - Mayıs 2019 tarihleri arasında elastofibroma dorsili toplam 84 hasta (12 erkek, 72 kadın; ort. yaş: 55.0±7.0 yıl; dağılım 43-74 yıl) retrospektif olarak incelendi. Baskın el, lokasyon, boyut, cinsiyet, yaş, meslek ve kitle takibi kaydedildi ve ameliyat öncesi ve sonrası Görsel Analog Ölçeği skorları not edildi.

Bulgular: Ortalama semptom süresi 9.5±7.8 (dağılım, 3-48) ay idi. Lezyonların 49'u (%58.3) sağda, 16'sı (%19) solda ve 19'u (%22.6) iki taraflı yerleşimli idi. Kitle boyutu ile yaş, semptom süresi ve komplikasyon gelişmesi arasında anlamlı bir fark yoktu (p=0.129). Kitle yerleşim yeri ile baskın kullanılan el arasındaki ilişki anlamlı idi (p=0.010). Ameliyat öncesi ve sonrası 90. günde Görsel Analog Ölçeği skorları anlamlı olarak değişiklik göstermiş olup (p<0.001), ağrı skorlarında anlamlı bir azalma izlendi.

Sonuç: Elastofibroma dorsinin cerrahi rezeksiyon sonrası klinik sonuçları iyidir. Bizim çalışmamızda, hastalarda elastofibroma dorsi anlamlı ölçüde baskın elde gözlemlendi.

Anahtar sözcükler: Göğüs duvarı tümörleri, elastofibroma dorsi, yumuşak doku tümörleri.

Elastofibroma dorsi (ELD) is a rare, slow-growing, and benign soft tissue tumor with solid, non-capsulated nature, located in the periscapular region of the posterior chest wall. Although ELD usually shows unilateral localization, it can be bilateral in 10% of

cases. It was first described by Jarvi and Saxen^[1] in 1961 and was typically reported to be located on the right side in most middle-aged female patients. The appearance of 81% pre-elastofibroma changes in the autopsy series suggests that ELD is an age-related

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process, and it has been reported that incidence of up to 2% in ELD computed tomography (CT) in elderly patients has been detected incidentally.^[2,3]

Elastofibroma dorsi consists of microscopically proliferated tumor cells of fibroblasts and accumulated abnormal elastic fibers. It is assumed that connective tissue degeneration is caused by repeated friction of the chest wall and tip of scapula, due to the use of the extremity.^[1] Therefore, it has usually been found in patients who frequently repeat occupational handwork. In the English literature, there is no clinical trial showing the exact relationship between recurrent mechanical trauma and ELD.

In this study, we aimed to investigate the correlation between the side of ELD formation and the dominant-hand of the patients we operated.

PATIENTS AND METHODS

This single-center, retrospective study was conducted at University of Health Sciences, Süreyyapaşa Chest Diseases and Thoracic Surgery Training and Research Hospital, Department of Thoracic Surgery between April 2007 and May 2018. A total of 84 patients with ELD (12 males, 72 females; mean age: 55.5 ± 7.0 years; range, 43 to 74 years) were included. Demographic data including age, sex, profession, mass size, duration of symptoms, dominant hand side, pre- and postoperative 90th day pain Visual Analog Scale (VAS) scores, and the recurrences occurred during follow-up were recorded. The diagnosis of ELD was confirmed by magnetic resonance imaging (MRI) in all patients (Figures 1a and 1b). The masses were

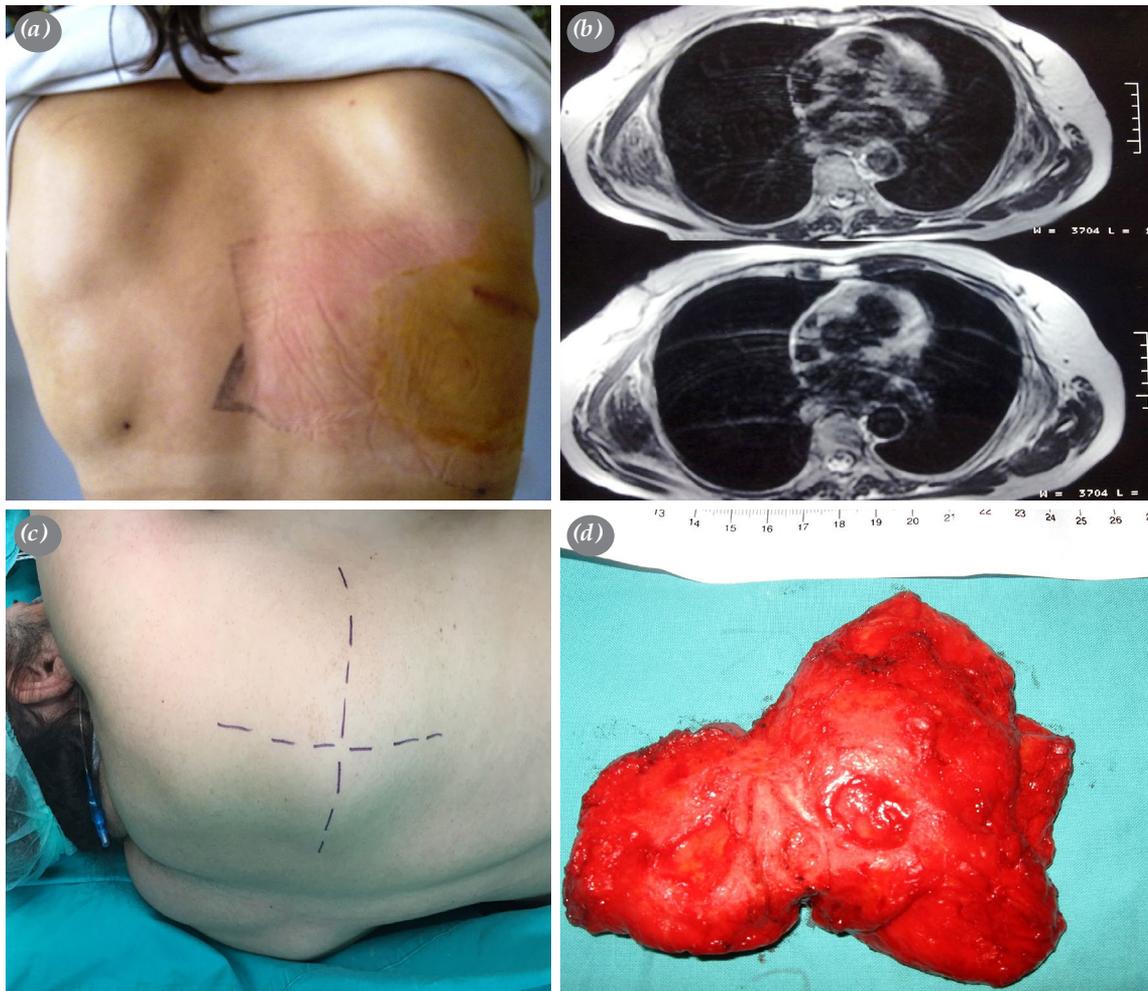


Figure 1. (a) The appearance of the ELD became visible in the subscapular with the abduction of the arm. (b) Magnetic resonance imaging section and (c) intraoperative view of ELD. (d) Macroscopic view of excised specimen.

ELD: Elastofibroma dorsi.

treated by total excision and no patient underwent preoperative incisional or needle-aspiration biopsy.

Surgical technique

All patients were operated under general anesthesia, in the posterolateral thoracotomy position, and patients with bilateral ELD in two sequential sessions with an interval of three weeks (Figure 1c). First, the latissimus dorsi muscle was separated by a transverse incision, then the solid-irregular shaped lesion attached to serratus anterior muscle and the periosteum of the ribs was excised totally. After careful bleeding control, a small drain was placed in the lobe and it was terminated when the drainage was ≤ 25 mL/day. Compressed dressings were used up to three days postoperatively to prevent possible seroma and hematoma complications.

Histopathological examinations were completed and confirmed by three pathologists.

Statistical analysis

Statistical analysis was performed using the IBM SPSS for Windows version 22.0 software (IBM Corp., Armonk, NY, USA). Descriptive data were expressed in mean \pm standard deviation (SD), median (min-max) or number and frequency, where applicable. The relationship between grouped variables was tested using the chi-square test. Repeated measurements and the difference between two independent groups were analyzed by paired group t-test, and Pearson correlation was used to identify differences between continuous variables. A *p* value of <0.05 was considered statistically significant.

Table 1. Patient characteristics

	n	%	Mean \pm SD	Range
Age (year)			55.5 \pm 7.0	43-74
Sex				
Male	12	15.3		
Female	72	85.7		
Side				
Bilateral	19	22.6		
Left	16	19		
Right	49	58.3		
Hand preferences				
Right-handed	68	81		
Left-handed	16	19		
Symptom				
Pain+palpable mass	52	61.9		
Pain	84	84		
Comorbidity				
No	39	46.4		
Diabetes	14	16.6		
Hypertension	35	41.6		
Complication				
Seroma	12	14.2		
Hematoma	3	3.5		
No complication	69	82.1		
Occupation				
Housewife	56	66.6		
Manual laborer	20	23.8		
Work at desk job	5	5.9		
Teacher	3	3.5		
Tumor size (mm)			77.6 \pm 16.7	50-130
Mean follow-up time (month)			32.5 \pm 20.1	
Duration of symptoms (month)			9.5 \pm 7.8	

SD: Standard deviation.

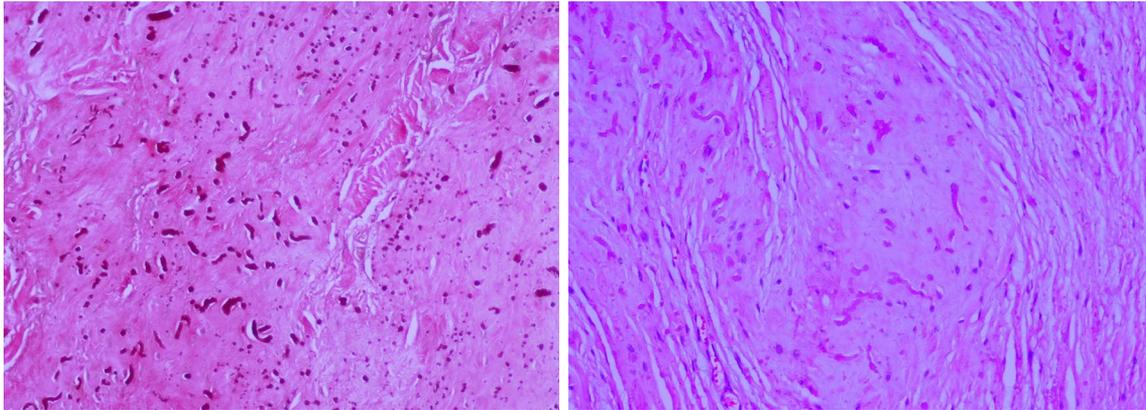


Figure 2. Microscopic views of the specimen (H&E, ×40).

RESULTS

Of a total of 84 patients, 56 (66.6%) were housewives, 20 (23.8%) were hand workers (ongoing or pre-made), five (5.9%) had desk jobs, and three (3.9%) were primary school teachers. All patients had pain that became evident with arm movements in the shoulder and back region (Table 1).

All patients presented with pain accompanying a mass, and it could be clearly seen with the abduction and elevation of the arm on dorso (Figure 1a). The diagnosis was confirmed by MRI in all patients. The mean duration of symptoms before applying to our clinic was 9.5 ± 7.8 (range, 3 to 48) months. The mean macroscopic dimension of the resected mass was 77.6 ± 29.8 mm (range, 50 to 130) mm (Figure 1d). Pathological examination revealed elastic and collagen fibers accompanying mature adipose tissue for all patients, consistent with ELD diagnosis (Figure 2).

The mean follow-up was 32.5 ± 20.1 (range, 4 to 120) months. Complications developed in 15 (17.9%) patients. In 12 patients who developed seroma, cure

was achieved with simple aspiration and pressed dressing, while three patients with hematoma were spontaneously resorbed with pressed dressings. No significant difference was found between patient age or lesion size and the complication development ($p=0.124$). During follow-up, no recurrence nor new onset contralateral ELD occurrence was observed.

Comparison of VAS scores at postoperative Day 0 (VAS0) and Day 90 (VAS90) scores showed that patients' pain complaints decreased significantly in the postoperative period ($p<0.001$).

The operated patients had 49 (58.3%) right-sided, 16 (22.6%) left-sided, and 19 (22.6%) bilaterally located lesions. A significant relation was found with the side of mass and the dominant hand ($p=0.010$) (Table 2). In addition, ELD was detected under the right scapula in 45 (66.2%) patients. In six (37.5%) of the patients using the left hand, the mass was observed on the left. When the statistical relationship between the side of the mass with larger size and the dominant hand was examined in patients with bilateral ELD, there was a significant relationship in the left-hand dominant

Table 2. Relationship between elastofibroma dorsi formation and dominant-hand side

	Dominant-hand				Total		<i>p</i>
	Right		Left		n	%	
Tumor side	n	%	n	%	n	%	
Right	45	66.2	4	25.0	49	58.3	9.233* 0.010
Left	10	14.7	6	37.5	16	19.0	
Bilateral	13	19.1	6	37.5	19	22.6	
Total	68	100.0	16	100.0	84	100.0	

* Chi-squared test.

group ($p=0.003$), whereas the right-handed ones were not significant ($p>0.05$).

DISCUSSION

The literature on ELD primarily consists of case reports and small series.^[4,27] Although its exact etiology is unknown, recurrent mechanical traumas in the subscapular region are reported to play an active role. It has been reported that ELD is more common in individuals with high repetitive upper extremity physical activity.^[4,14] Elastic degeneration, reactive hyperproliferation in collagen or fibroblastic tissue develops in this region due to recurrent micro-traumas between the scapula and the chest wall.^[4,15] In our series, we attempted to reveal the accuracy of this mechanical theory. We investigated whether ELD formation was more dominant on the hand side, which is used to determine the location of repetitive micro-traumas in the etiology of arm movements, particularly hand workers. The fact that the lesion of patients we operated was usually on the same side with their dominant hand used in daily life supported our theory (Table 2). In the studies conducted, the dominant hand is more in the Turkish population.^[16]

In addition to this mechanical theory, there are studies in the literature containing two controversial evidences supporting genetic predisposition theory.^[14,17] In the largest series in literature of 170 cases, about 38% reported familiar predisposition (not a specific study of genetic status).^[14] The second genetic theory is that it occurs due to varying degrees of gene's hypo- or hyper-expression on different chromosomes.^[18,19] Some authors have also explained the formation of ELD due to vascular insufficiency and relative ischemia in the scapular area. This pathogenesis theory has not been extensively addressed and has only been reported as a reference hypothesis so far.^[20] Considering the characteristic features of ELD, it is evidence supporting that it is caused by mechanical repetitive micro injuries rather than genetic predisposition, rather than being seen more frequently in individuals over the age of 50 years.^[21,22] In our cohort, we did not encounter any patient with a family medical history with previous ELD, in contrast with this theory for our study.

Although ELD is known to be a very rare benign soft tissue tumor, its actual incidence may be higher. The CT examinations taken in the normal population reveal an ELD incidence of 2%.^[3] In autopsy series, ELD was observed in 11.2% in males and 24.4% in females over 55 years old.^[4] In our series, the number of women (85.7%) with ELD was higher than men; mostly were reported to be housewives (66.6%, $n=56$).

The incidence of bilateral EDL was reported to be between 12 and 73%,^[3,5] and it was 22.6% in our series.

Pain and mass formation in the subscapular region are the most common symptoms. In the series of 76 cases, pain (93%), functional restriction (51.3%), and mass appearance during abduction of the arm (31%) were the commonest complaints of patients with ELD.^[6] In our series, there were pain complaints in all 84 cases, and in 52 (61.9%) cases, there was also a mass that accompanied the pain (Table 1). A similar literature study reported that the VAS score of the patients decreased from 4.6 to 2.4 at 4.6 years of follow-up.^[7] When we compared the pre- and postoperative VAS scores, we observed that pain decreased after the removal of the mass significantly ($p<0.001$).

Due to common clinical symptoms, ELD can get misdiagnoses such as rotator cuff syndrome or cervical vertebrae problems. Some cases may not be diagnosed, unless they are over 5 cm in CT or MRI.^[8]

Magnetic resonance imaging is the best tool to confirm the diagnosis (Figure 1b). We observed that patients who did not complain of mass, but just with back pain complaints were evaluated by orthopedics and physical rehabilitation clinics many times beforehand and could not be diagnosed correctly for a long time. Fibrous tissue shows a low signal intensity in the T1 and T2 sequences, while fat tissue shows high signal intensity in the T1 sequence. Adipose tissue appears as lines in fibrous tissue parallel to the chest wall.^[9] It is used in characteristic cases as a diagnostic method in ultrasound (US).^[10] In studies using positron emission tomography (PET)/CT, the mean standard uptake value (SUV) of 18F-fluorodeoxyglucose has been shown to be 2.29.^[11] Blumenkrantz et al.^[12] reported that PET/CT detected ELD in 1.66% of patients. Only two of our patients had PET/CT scans taken simultaneously for other reasons. The SUV of these patients were 2.35 and 1.99, respectively. Since no malignant transformation of ELD, except for only one case, was reported of high spindle cell sarcoma and ELD together in the current literature,^[13] we did not perform biopsy in any patient preoperatively.

None of the patients we operated in our series had a family history. The mean age of the patients was 55.5 ± 7.0 years. In this study, no significant relationship was found between mass size and age ($p=0.124$). Although indications for surgery are still controversial, resection is usually recommended in symptomatic cases over 5 cm. Some authors argue that surgical

resection is necessary for pathological confirmation, whether symptomatic or not.^[23,24] All of the patients in this series were patients who presented with pain complaints, and we found that, particularly after the surgery, pain decreased and daily activities increased, significantly.

Recurrence rates between 0.06 and 4.5% are reported in the literature.^[14,25] In our series, we performed a surgical re-resection due to recurrence in a patient operated by a general surgeon 15 years ago. No ipsilateral- or contralateral-occurrence/recurrence was seen in any patients during our mean duration of 32.5±20.1 months, postoperatively.

The most frequently reported complications after ELD resection are hematoma and seroma formation, as high as 40 to 50%, directly proportional with increasing tumor size.^[14,26,27] There is no consensus on postoperative rehabilitation, but one week of immobilization is recommended.^[26] We did not restrict any patient's mobilization, but we applied compression dressing, to prevent early postoperative hematoma formation, and resulted in 18% incidence of hematoma or seroma within the first 30 days. In patients who developed seroma after discharge, we achieved treatment with needle aspiration and pressurized dressing.

The main limitations of this study are its single-center and retrospective design. However, comparatively high cohort volume and long follow-up duration features were the main strengths. To the best of our knowledge, our study is the first study to reveal the relationship between the dominant hand used and the development of the tumor, and is also the second largest series in the English literature,^[14] as well as the largest case series in the thoracic surgery literature, proving the benefits of surgical treatment for patients suffering severe pain.

In conclusion, elastofibroma dorsi, which is a rare, slow growing soft tissue tumor located in the subacapular region, has good clinical results after surgical resection. In our series, the patients had significantly more elastofibroma dorsi on their dominant-hand side. However, we believe that it is appropriate to examine larger case series to reveal the definitive etiological relationship.

Ethics Committee Approval: The study protocol was approved by the Süreyyapaşa Chest Diseases and Thoracic Surgery Training and Research Hospital Ethics Committee (Date/no: 06.05.2021/116.2017.R-219). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Patient Consent for Publication: A written informed consent was obtained from each patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Conception - S.K.M.; Design - S.E.; Data collection and processing - S.K.M., S.E.; Analysis and interpretation - S.K.M., S.E.; Final approval - S.K.M., S.E.

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