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Clinical features, imaging findings, treatment aspects of elastofibroma dorsi and long-term outcomes after surgical resection

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Abstract. – OBJECTIVE: We aim to present clinical features, imaging findings, treatment aspects of the elastofibroma dorsi (ED), which is a benign tumor arising from connective tissue at the scapular region, and long-term outcomes after surgical resection.

PATIENTS AND METHODS: We evaluated retrospectively 82 patients (55 females, 27 males; mean age, 60 years; age range, 23-78 years) with ED who underwent surgery between January 1994 and May 2014; subsequently all patients were invited for follow-up, which consisted of physical and US examinations.

RESULTS: Subscapular location was almost constant (79/82 patients). Right, left and bilateral location was noted in 39, 28 and 15 cases, respectively. 52/82 patients were symptomatic. The diagnosis was made on physical examination and imaging studies: 49 ultrasound, 43 computed tomography and 54 magnetic resonance examinations were performed overall. Surgical treatment consisted in marginal excision; in all cases diagnosis was confirmed by histological examination. The mean hospitalization was 3 days, with minor complications. Out of the 82 patients, only 25 gave their consent to follow-up; mean time passed after surgery was 64.7 months; 1 case of local recurrence was suspected by ultrasound and, then, confirmed by magnetic resonance imaging.

CONCLUSIONS: In our series, clinical features and imaging findings of ED are consistent with current evidence; however, results of our follow-up group marks a difference from the literature, according to which there is no evidence of local recurrence after complete resection. Diagnosis of ED is based on clinical and imaging features; treatment is surgical, especially in symptomatic cases. Prolonging the clinical and US follow-up period may be useful in identifying local recurrence.

Key Words
Elastofibroma, Imaging findings, Diagnosis, Treatment, Follow-up.

Introduction

Elastofibroma dorsi (ED) is a slow-growing benign connective tissue tumor of the thoracic wall with no malignant potential. The lesion usually arises beneath the rhomboid major and latissimus dorsi muscles adjacent to the inferior angle of the scapula and can be bilateral in about 25% of cases. Very rarely (1% of cases), elastofibroma can occur at other sites such as near the greater trochanter and the ischial tuberosity[1-6]. It is more common in females after the fifth decade and it usually manifests as a palpable mass on the mobilization of the shoulder, with or without pain (5). Although ED is regarded as an uncommon tumor, there are reports of a prevalence of 2% in people over 60 years, while an autopsy study found a prevalence of 24% in females and 11% in males[6].

Despite the lack of a universally shared diagnostic algorithm, the diagnosis is primarily based on clinical presentation and imaging features. On physical examination, the lesion is usually well-circumscribed and mobile, although it may be difficult to precisely discern it from surrounding structures[3,7]. Ultrasound (US), computed tomography (CT) and magnetic resonance (MR) imaging are used as complementary examinations to confirm the clinical suspicion of ED[3,8]. The US typically shows an abnormal mass, generally in the subscapular region, with an alternating pattern of hyperechogenic and hypoechogenic lines that are

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roughly parallel to the chest wall[9]. At CT, ED may be seen as a homogeneous mass with a density inferior to that of muscles with scattered areas of decreased attenuation, suggesting the presence of fat within the lesion[5]. MR imaging has a higher diagnostic confidence for ED when compared to US and CT, especially with T1- and T2-weighted sequences. In fact on MR imaging, ED is typically a well-defined, heterogeneous soft-tissue mass with a signal intensity similar to that of skeletal muscle, frequently with intermixed linear or curvilinear streaks of fat signal intensity. Contrast administration is not necessary to characterize the lesion when typical findings are present at MR imaging, as contrast enhancement is fairly variable, ranging from a mild to avid enhancement[6]. Therefore, specific clinical features, including subscapular location and patient age at presentation, as well as typical imaging findings at US/CT/RM, such as entrapped fat within a circumscribed fibrous mass, can clinch a diagnosis with a high degree of confidence, particularly when bilateral lesions have been identified. Most of the recent publications[7-9] indicates that biopsy is unnecessary when imaging findings are sufficiently typical and interpreted in the light of appropriate clinical findings. The biopsy should be conducted when imaging findings are atypical or in the case of fast-growing lesions; in fact, the differential diagnosis includes benign and malignant lesions such as hematoma, hemangioma, aggressive fibromatosis, lipoma, malignant fibrous histiocytoma, fibrosarcoma, liposarcoma or metastases[1,8]. Local excision is the treatment of choice, especially for symptomatic ED, with few post-operative complications. According to literature, there is no evidence of recurrence of disease in patients with complete marginal resection; rare recurrences are probably due to incomplete excision[1-9].

The aim of this work is to present clinical features, imaging finding, treatment aspects of the ED and long-term outcomes after its resection.

Patients and Methods

We retrospectively reviewed 82 patients (55 females, 27 males; mean age, 60 years; age range, 23-78 years) who underwent surgical resection of ED at different Departments of our institution (Thoracic Surgery, General Surgery, Orthopedics, Plastic Surgery) between January 1994 and May 2014; furthermore, we assessed pre- and post-operative evaluations and follow-up records.

Pre-operative evaluation

Medical records from our hospital as well as from other hospitals were reviewed in order to determine location, symptoms, imaging findings and treatment of lesions.

Post-operative evaluation

Details regarding histology and post-operative courses were obtained retrospectively from in-house clinical records. Factors that may affect post-operative complications like tumor size and placement of post-surgical wound drainage were assessed.

Follow-up

All 82 patients, 5 of which with bilaterally resected ED, were telephonically invited for follow-up, which consisted of physical and US examinations; 25 patients gave their consent. During physical examination we evaluated: the presence of a palpable lump along the surgical scar; the presence of pain in the site of intervention with particular attention to its duration, its intensity according to the Visual Analogue Scale (VAS, 0 represented no pain and 10 represented excruciating pain) and its eventual relation to active mobilization of the shoulder. All 25 patients underwent US with Esaote apparatus (MyLab™ClassC, Genoa, Italy) using linear transducers (5-13MHz) to obtain sagittal and transverse scanning planes of the site of intervention. In patients who referred pain during active mobilization at physical examination, US study was extended to the region of the shoulder, in order to exclude concomitant rotator cuff disorders. In the event of suspected local recurrence of ED, diagnosis would have to be confirmed by MR imaging, performed with Signa 1.5-T unit (GE Medical Systems, Milwaukee, WI, USA) with multiplanar T1-weighted spin-echo sequences (TE 10, TR 500), T2-weighted fast spin echo sequences (TE 85, TR 2,800) and short-tau inversion recovery (STIR) sequences (TE 14, TR 4,800) with slice thickness of 4 mm. The contrast agent used was gadolinium chelates (MultiHance, Bracco, Colleretto Giacosa, Turin, Italy) injected at 0.1 mg/kg using an automatic power injector to administer a single bolus at a rate of 1 ml/s, followed by acquisition of multiplanar T1-weighted spin-echo sequences with fat saturation (TE 10, TR 500). We used high-resolution sequences with 256×256 matrix and variable field of view (FOV). The present study was planned and conducted in accordance with the Ethical standards of the responsible Committee on human experimentation and conformed to the principles of the Declaration of Helsinki.

Results

Pre-operative evaluation

ED was situated in the subscapular region in 79 patients (96.3%) and in the parascapular region in 3 patients (3.7%). The tumor was located on the right side in 39 cases (47.5%), on the left side in 28 cases (34.2%), and bilaterally in 15 cases (18.3%). 52 patients (63.4%) presented symptoms associated with the tumor, including subscapular swelling, pain around the scapula and snapping of the scapula during movement of the shoulder. The time from the onset of symptoms to the discovery of the lesion ranged between 30 days and 93 months. In 30 asymptomatic patients (36.6%) the mass was incidentally discovered during radiological examinations performed for different medical conditions. Imaging studies were available in all 82 patients: 49 US, 43 CT and 54 MR imaging studies were performed overall. At all imaging modalities, tumors appeared as poorly circumscribed heterogeneous soft tissue masses with a consistency similar to that of skeletal muscle interlaced with strands of fat (Figure 1). Therefore, physical and imaging examinations showed typical findings of ED in all cases and marginal resection of the tumor without pre-surgical biopsy was performed in all patients; in only 3 cases (3.6%), unnecessary biopsies were executed in other hospitals prior to referral to our institution. In 15 cases of bilateral ED sequential surgery was proposed, in order to allow free activity of the non-operated shoulder. In this subgroup, only 8 patients underwent sequential surgery: in fact, in 2 cases synchronous bilateral resection was performed, while 5 patients refused second intervention.

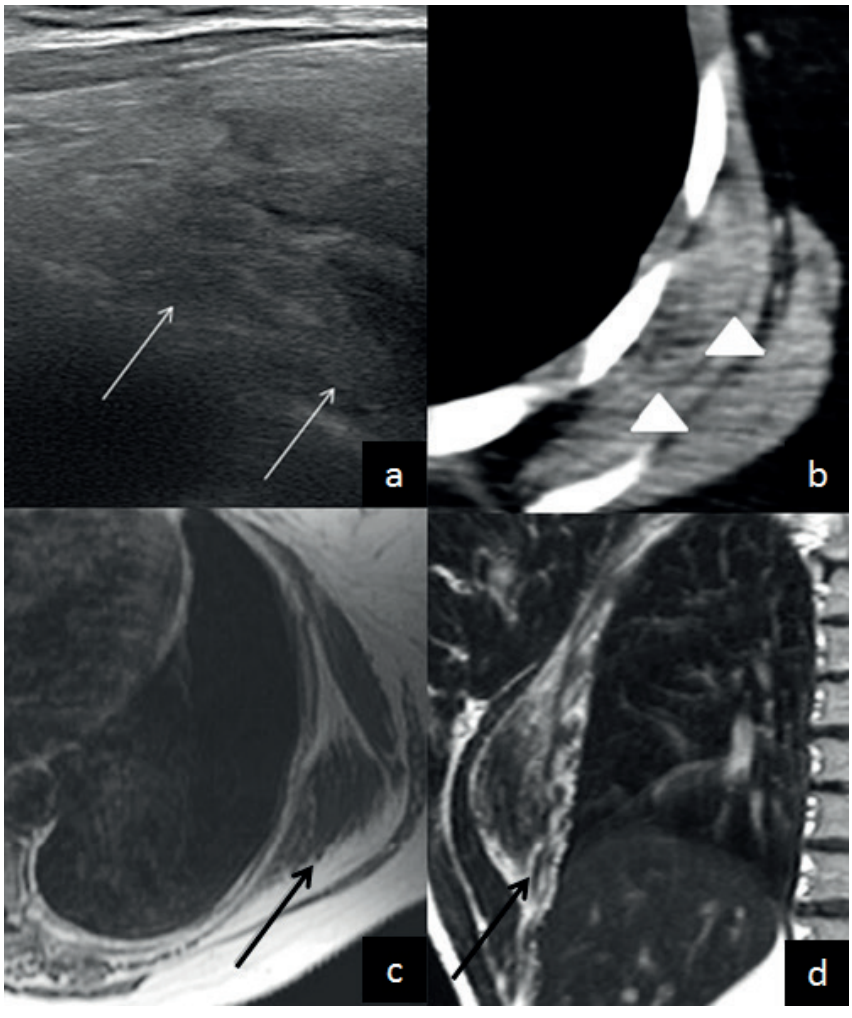

Figure 1. Subscapular ED in different patients on imaging studies. **a**, Sagittal US scan shows an ill-circumscribed soft tissue mass with interlaced hypoechoic and hyperechoic strands corresponding to fibroblastic and fat tissue *(arrows)*. **b**, Axial post-contrast CT scan shows a subscapular mass with a density similar to that of skeletal muscle with moderate enhancement *(arrowheads)*. **c**, Axial T1- and (**d**) coronal T2-weighted MR images show a poorly circumscribed, heterogeneous mass deep to the muscles and adjacent to the chest wall; the lesion has intermediate signal intensity similar to that of adjacent skeletal muscle, with interspersed linear areas of increased signal intensity similar to that of fat *(black arrows)*.

Post-operative evaluation

A total of 92 tumors were surgically removed; 90 ED were un-capsulated (97.8%) while 2 were capsulated (2.2%). The maximum mean diameter of the lesions was 7.7 cm (range, 1.5-16.5 cm). The histological examination of all 92 lesions confirmed a hypocellular mass with large eosinophilic elastic fibers and collagen network of mature adipose cells compatible with ED, which was completely resected both macroscopically and microscopically. Patients were discharged between the 1st and the 8th post-operative day, with a mean hospitalization of 3 days. Minor post-operative complications were identified in 13/92 cases (14.1%), in particular: 7 patients developed a seroma; 6 patients developed a post-operative hematoma. Due to the relative incidence of post-operative seroma and hematoma, factors that may affect post-operative complications such as tumor size and placement of post-operative wound drainage were investigated. The comparison between the group with post-operative complications and the group without post-operative complications showed a larger mean tumor diameter (8.9 cm *vs.* 7.6 cm respectively) and a lower percentage of post-operative wound drainage placement (84.6% *vs.* 87.3% respectively) in the first group.

Follow-up

The mean follow-up period was 64.7 months (range, 8-173 months). Out of the 82 patients, only 25 gave their consent to follow-up; 5 of these underwent bilateral surgery. Therefore, we performed a total of 30 physical and US examinations. During the physical examination, none of the patients presented a palpable lump at the site of intervention. In 21/30 cases (70%) we reported the absence of pathological symptoms along the surgical scar and a normal US finding. In 6/30 cases (20%) we noted chronic pain unrelated to the active mobilization of the shoulder (pain lasting more than 12 weeks; VAS range, 4-7). Among this subgroup, at the US we noted a minimal thickening of soft tissue along the surgical scar in 5 cases and a suspected disease recurrence in 1 case, which was confirmed later by the MR imaging (Figures 2, 3). In 3/30 cases (10%) we reported chronic pain during active mobilization of the shoulder (pain lasting more than 12 weeks; VAS range, 2-4) due to rotator cuff disorders shown by the US.

Table I summarizes clinical characteristics of all patients considered in the analysis.

Discussion

ED is a rare, benign, slow-growing soft tissue tumor that primarily affects the elderly, with a mean age of about 60 years at diagnosis. ED is present more frequently in females than in males (females:males ratio, 4:1). The most frequent location is the subscapular region; in few cases, it can be found in the parascapular region or other sites of the musculoskeletal system. It occurs predominantly on the right scapular side, but it is found bilaterally in about 25% of cases[6,8,10]. Moreover, ED tends to be asymptomatic in more than half of the cases[3,5,10-12]. In our study group, epidemiological features were in accordance with those in literature, except for the females:males ratio (2:1) and for the proportion of asymptomatic patients (36.6%). Diagnosis is based primarily on physical examination and imaging studies, such as US, CT and MR imaging, that are used as complementary examinations to confirm the clinical suspicion (Figure 1). US is usually the first-line imaging modality performed (non-invasive, quick and inexpensive), identifying the lesion as an oval mass, with ill-defined margins, fixed to the deep costal plane. The limitation of US is represented by the patient's body build with possible deep artifacts that can falsify echogenicity of the lesion and its margins with a possible underestimation of its real size[10]. In our series, US was carried out in 49 patients showing an abnormal mass of tissue in the subscapular/parascapular region with a typical alternating pattern of hyperechogenic and hypoechogenic lines parallel to the chest wall. In our experience when the clinical and US data are typical for ED, CT or MR imaging are not necessary[3,5,10]; CT or MR imaging should be performed in cases of atypical findings at clinical and US examination or in the pre-surgical evaluation. MR imaging is the non-invasive imaging modality with the greatest sensitivity and specificity in detecting ED because it shows a circumscribed mass with an alternating pattern of fibrous and fatty tissues. On both T1- and T2-weighted sequences fibrous tissue produces low-intensity signal identical to muscular tissue, while the fatty tissue is seen as a high-intensity signal on T1-weighted sequences, as an intermediate signal on T2-weighted sequences and as a low-intensity signal on STIR sequences. Gadolinium related contrast enhancement is heterogeneous. CT is less sensitive than MR imaging in visualizing the fat component, so that ED may be seen as a homogeneous mass with a density inferior to that

Table I. Summary of clinical characteristics of all patients considered in pre- and post-operative evaluations of our analysis, and, clinical characteristics of patients considered at follow-up. pts = patients.

Pre-operative evaluation of 82 patients with ED (55 females, 27 males; mean age, 60 years)			
Scapular location	Subscapular 79 pts (96.3%)	Parascapular 3 pts (3.7%)	—
Site	Right 39 pts (47.5%)	Left 28 pts (34.2%)	Bilateral 15 pts (18.3%)
Symptomatic pts	Yes 52 pts (63.4%)	No 30 pts (36.6%)	—
Imaging studies	US 49	CT 43	MR 54
Pre-surgical biopsy	Yes 3 pts (3.6%)	No 79 pts (96.3%)	—
Surgical treatment	Yes 82 pts (100%)	No 0 pts (0%)	—
Bilateral surgery (15/82 pts)	Sequential surgery 8 pts	Synchronous surgery 2 pts	No second intervention 5 pts
Post-operative evaluation of 92 ED			
Cases of capsulated/un-capsulated ED		Capsulated 2 cases (2.2%)	Un-capsulated 90 cases (97.8%)
Mean diameter of ED		7.7 cm	—
Cases with histological confirmation of ED		Confirmed 92 cases (100%)	Not confirmed 0 cases (0%)
Cases of ED completely resected		Yes 92 cases (100%)	No 0 cases (0%)
Mean of days of hospitalization		3 days	—
Cases with post-operative complications		Yes 13 cases (14.1%)	No 79 cases (85.9%)
Follow-up of 30 ED			
Mean period passed after intervention	64.7 months	—	—
Cases with palpable lump in the site of intervention		Yes 0 cases (0%)	No 30 cases (100%)
Cases with chronic pain in the site of intervention	Yes, unrelated to active mobilization 6 cases (20%)	Yes, related to active mobilization 3 cases (10%)	No 21 cases (70%)

of muscles. Thus, when necessary, MR imaging study should be considered the investigation of choice as complementary to an examination of US[3,5,10,12]. In our series, CT and MR imaging were performed respectively in 43 and 54 cases for pre-surgical evaluation or for different medical conditions in which ED was incidentally discovered. In all patients imaging features of ED were so characteristic that pre-surgical biopsy was not performed, except in 3 patients in which biopsy was performed in other hospitals prior to referral to our institution[13,14]. Generally, conservative treatment is recommended in elderly and asymptomatic patients; in fact, to date, malignant transformation has never been reported[5,7]. On the other hand, the indication for surgery depends on the severity of symptoms and patient preference. Marginal excision is sufficient, and surgery may bring dramatic relief of troublesome symptoms. However, there are several reports[7,8,13] showing an incidence of minor post-operative complications, such as seroma or hematoma, ranging from 11.8% to 35.1%. In all patients of our cohort, complete surgical resection was performed, both in symptomatic patients (the majority of cases) as well as in asymptomatic ones, due to patients' aesthetic desires; minor post-operative complications were identified in 14.1% cases, rates similar to those identified in the literature[7,8,13]. Our investigation of factors that may influence post-operative outcomes[7,8,13-19] suggests the use of post-operative wound drainage in order to prevent complications, especially in the case of ED with large tumor diameter. According to the limited literature available[7,8,13,14,20,21], there have been no reports of residual symptoms or local recurrence after

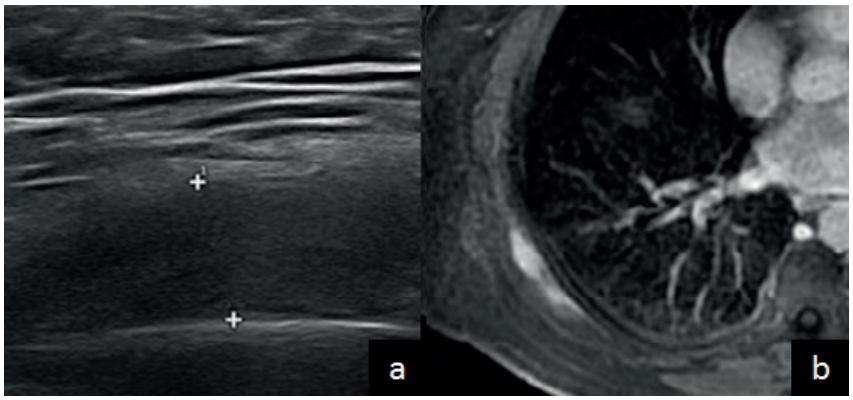

Figure 2. Local recurrence of ED in a 56-years-old woman. **a**, Sagittal US scan shows an inhomogeneous hypoechogenic subscapular mass deep to the surgical scar. **b**, Axial post-contrast T1-weighted fat-suppressed MR image shows a subscapular mass with moderate enhancement after intravenous contrast administration.

complete marginal excision of ED. However, in our follow-up group, we found a subgroup of 6/30 cases (20%) with mild/moderate chronic pain along the surgical scar not referable to a single nerve distribution, unrelated to active mobilization of the shoulder; among this subgroup, we noted 1 recurrence at 76 months (recurrence rate, 3.3%). At US, ED recurrence was strongly suggested by an inhomogeneous hypoechogenic subscapular mass; however, post-operative scar or granulation tissue couldn't be excluded in the different diagnosis; therefore, MR imaging was subsequently performed. Thus, MR imaging confirmed ED recurrence showing low signal on T1- and T2-weighted sequences from fibrous components mixed with high signal intensity on T1- and T2-weighted sequences from fat components; ED showed enhancement after intravenous contrast administration (Figure 2). This differed from post-operative fibrosis usually characterized by low signal intensity on both T1- and T2-weighted sequences and with no or poor post-contrast enhancement. In the other 5 cases of the follow-up subgroup, characterized by a mild/moderate chronic pain along the surgical scar not referable to a single nerve distribution unrelated to

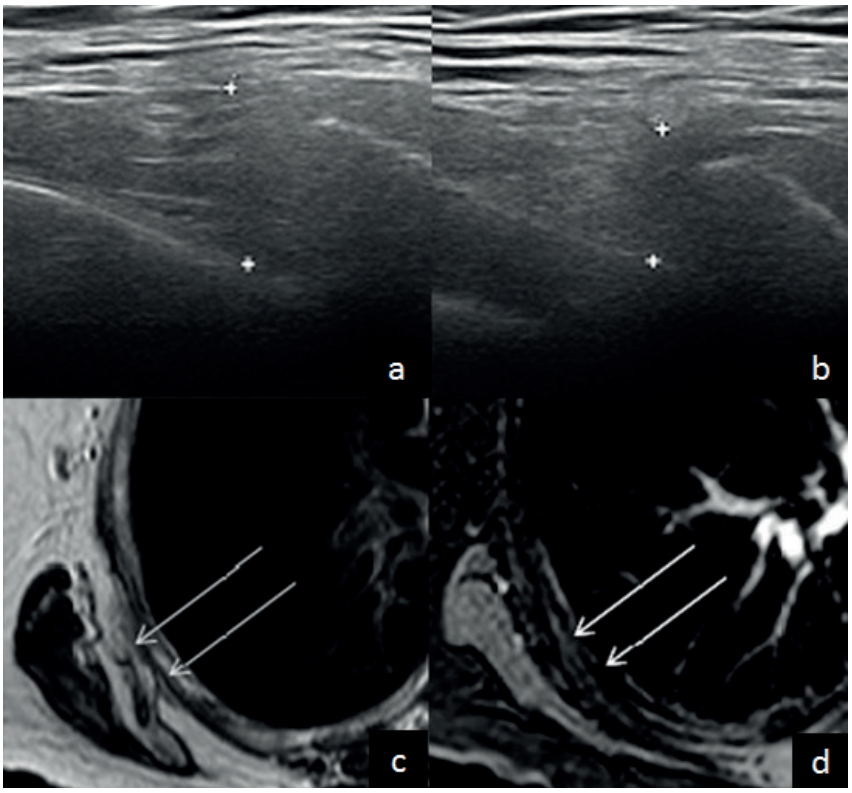

Figure 3. Post-operative fibrosis in a 46-years-old woman with right ED surgically resected. (**a**, **b**,) Sagittal US images show minimal thickening of soft tissue deep to the surgical scar compared to the normal contralateral side. **c**, Axial T1- and (**D**) axial post-contrast T1-weighted fat-suppressed MR images show post-operative fibrosis characterized by low signal intensity and no enhancement following intravenous contrast enhancement *(arrows)*.

active mobilization of the shoulder, US examination showed no pathologic conditions, except for minimal thickening of soft tissue along the surgical scar due to post-operative fibrosis (Figure 3). According to the literature, chronic pain experienced by these patients might be related to injury of the nociceptive system, which occurs during a surgical procedure, and might benefit from treatment by a specialist in pain therapy[22,23]. Our study has some limitations: it is a retrospective study with a moderate sample size; further studies are necessary to define long-term outcomes and follow-up of surgically resected ED. However, according to our work, before surgical resection of ED, patients should be informed of the possible risks of local recurrence, even in cases of complete excision, and of the possibility of injury to the nociceptive system at the surgical site. Therefore, US could be a non-invasive, quick and inexpensive follow-up examination in early identification of these long-term complications, especially in patients with chronic pain along the surgical scar after marginal resection of ED.

Conclusions

ED is an uncommon benign tumor usually seen in the subscapular region after the fifth decade of life with a female predilection. Diagnosis of ED is based on clinical and imaging features. Treatment is controversial; complete surgical resection should be performed if the lesion is symptomatic. Prolonging the clinical and US follow-up period may be useful in identifying local recurrences and other post-surgical injuries.

Conflict of Interests

The Authors declare that they have no conflict of interests.

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