

Chest Elastofibroma – Elastofibroma Dorsi

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Key words:

- Chest Elastofibroma

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SUMMARY

Chest wall primary tumors include a broad spectrum of benign and malignant tumors. Tumor biopsy is essential to set diagnosis. This rule does not apply to the elastofibroma dorsi. Elastofibroma is an unusual, well defined tumor of soft tissues typically located in the peri-scapular region, composed of characteristic abnormal elastophilic fibers and adipose tissue. It affects mainly elderly people and especially women. In magnetic resonance imaging and computed tomography elastofibroma presents a characteristic layered pattern of fatty and fibrous tissue. The typical location and the characteristic imaging pattern set the diagnosis of elastofibroma without the need for biopsy. Biopsy is required in atypical cases to exclude malignancy, especially sarcoma. In asymptomatic cases, simple monitoring is sufficient. Surgical excision is recommended for serious symptoms and diagnostic doubt. The prognosis is excellent. No case of malignant transformation has been described.

Pneumon 2017, 30(3):157-164.

INTRODUCTION

Chest wall is a site of location for primary and metastatic tumors. Chest wall primary tumors represent 1-2% of all primary tumors¹ and include benign and malignant tumors originating from the bones, cartilage and soft tissues (muscles, vessels, nerves) of the chest wall, and some haematological diseases².

Chest wall benign tumors consist the 56% (23/41) of the cases and include enchondroma, fibrous dysplasia, neurilemmoma, osteochondroma, granular cell tumor, fibroma, lipoma, fibrolipoma, eosinophilic granuloma, aneurysmal bone cyst. Chest wall malignant primary tumors account for 46% (18/41) of the cases and include plasmacytoma, chondrosarcoma, osteosarcoma, fibrosarcoma, desmoid tumor, leiomyosarcoma, malignant fibrous histiocytoma, tendon sheath sarcoma, hemangiosarcoma, and neurofibrosarcoma. The most common are the cartilaginous tumors (chondroma and chondrosarcoma) and then those derived of the soft tissue (fibromas, lipomas, neurogenic tumors)¹.

Consequently, the presence of a tumor in the CWI is a diagnostic chal-

lenge for the clinician. The likelihood of malignancy and especially of sarcoma forces the clinician to consider all chest wall tumors as malignant until evidence of the opposite¹. In this context tumor biopsy becomes necessary for tumor identification.

This rule does not apply fully to elastofibroma³. This benign tumor has in many cases typical clinical and imaging features that allow for safe diagnosis⁴, avoiding further biopsy investigation or even surgical excision⁵.

The presentation of the clinical and imaging characteristics of elastofibroma is the purpose of this review.

DEFINITION

Elastofibroma or elastofibroma dorsi (ED) is a benign, slowly growing tumour of soft tissue of subscapular area, composed of characteristic abnormal elastophilic fibers and adipose tissue. It was first described by Järvi and Saxen in 1959 at the 12th Congress of Scandinavian Pathologists, who then published their work in 1961³.

The 2002 World Health Organization (WHO) soft tissue tumor classification ranked the ED in the benign and myoinoblastic soft tissue tumors of the peri-scapular region⁶.

Location

ED is typically located between the lower corner of the scapula and the posterior CW. It may be connected to the periosteum of the ribs but without invading it or other adjacent structures⁷.

Elastofibroma may be unilateral or bilateral⁸. Bilateral location is more frequent with a reported frequency in different series at 58.62% (17/29)⁹, 66.27% (112/169)⁸, 75% (6/8)¹⁰, and 100% (9/9)⁷ of cases.

Unilateral elastofibromas are more commonly located in the right chest wall with a reported frequency of 61% (35/57) of cases⁸.

Size - Symmetry

The diameter of surgically excised tumours ranges from 2 cm to more than 15 cm^{8,11}, while in the computed tomography (CT), very small elastofibromas with diameter 1×1.5 cm⁵ and 2×0.5×2 cm¹² have been described.

The diameter of the larger palpable tumours is similar to physical examination and CT¹³. However, elastofibromas with diameter of less than 3 cm are difficult to be found in physical examination and CT⁷.

Bilateral elastofibromas may be symmetrical in size

and location¹⁴ or asymmetric^{5,7}, and many times asynchronous¹⁰.

Unusual location of elastofibromas

Elastofibromas are typically located at the periscapular region. However, a growing number of publications in the literature have described elastofibromas located at various anatomical sites, mainly the musculoskeletal system, and more rarely the visceral organs and the gastrointestinal tract¹⁵.

Unusual locations of elastofibromas have been described in eye¹⁶, face¹⁵, intraspinal space¹⁷, mediastinum¹⁸, tricuspid valve¹⁹, hand²⁰, foot²¹, axilla and inguinal region²², infraolecranon regions⁸, ischial tuberosity⁸, deltoid²³, greater trochanter¹⁶, posterior elbow¹⁶, pancreas¹⁵, stomach²⁴, intestine²⁵, sigmoid colon²⁶, and greater omentum²⁷.

These locations may be single or multiple, coexisting with the typical location in the sub-scapular region. Nagamine N et al⁸ in a clinical-pathology study involving 170 cases of ED described 27 cases in which the tumour was located in both sub-scapular areas and in addition in the infraolecranon regions, a case with seven different anatomical locations (sub-scapular, bilateral thoracic wall, and both infraolecranon areas), a case with four different anatomical locations (sub-scapular on both sides, left infraolecranon, and right ischial tuberosity) and a case with tumour in both sub-scapular areas and in the right ischial tuberosity. One patient had a single tumor located only in the olecranon. Shimizu S et al²⁸ described a case which presented 15 separate elastofibromas. The location of the masses included the classic sub-scapular sites as well as buttocks and upper limbs.

EPIDEMIOLOGY

Frequency

Although ED is considered as a rare entity, its exact frequency is not known. Significant differences exist in the described incidence of ED between clinical and pathology studies.

Pauline H. Go et al⁵ in a study based on data from 14 series of elastofibroma cases published between 1980 – 2009 described 330 symptomatic cases of ED. Brandser et al¹² in a CT study involving 258 asymptomatic patients over 60 years of age found five elastofibromas in 4 patients (prevalence of 2%). Blumenkrantz et al⁹ described 29 cases of ED in a total of 1,751 patients subjected to PET-CT with 18F-fluorodeoxyglucose (FDG), (1.66%). Review of Ortho-

pedic Oncology data from the Royal Orthopaedic Hospital in Birmingham, UK, revealed 15 cases of elastofibromas in a total of 17,500 cases (0.086%) in patients >20 years of age¹⁶. Nagano S et al²⁹, in an analysis of the Japanese Soft Tissue Tumor Registry database, found 130 cases of ED in a total of 12,557 cases with soft tissue volume (1%).

In contrast, autopsy studies have described a much higher incidence of ED.

Jarvi and Lansimies³⁰ in a series of 235 autopsies, found changes in the subcapsular thoracic fascia similar to ED in 39 cases. In people over 55 years, the frequency was 24.4 per cent in females (29 of 119) and 11.2 per cent in males (10 of 89).

Giebel et al³¹ in a study of 100 autopsies revealed 13 elderly patients with elastofibroma. Pre-elastofibroma-like morphological changes (e.g. few or many degenerated elastic fibres) were observed in 81% of the autopsies.

The differences in the reported incidence between clinical and autopsy studies suggest that the actual incidence of ED is higher than that described in the clinical studies. This is also supported by the fact of incidental discover of asymptomatic elastofibromas^{5,7,10,12}.

The absence of symptoms and the small size are obvious causes for the escape of the elastofibromas⁵.

Racial - Age Distribution

ED affects mainly elderly patients. The average age at diagnosis in different series has been described in 62 years (range 6-94 years)⁵, 63.7 years (range 46 - 79 years)²², 54.9 years (range 44 - 62 years)¹⁰, 61.1 years (range 38-78 years)¹¹, 65 years (range 48-72 years)⁷ and 68.4 years (range 51-79 years)¹⁶. In contrast, ED is rare in children and adolescents³¹.

Elastofibroma affects predominantly women. The female/male ratio has been described in 2.2:1 (11 women/5 men)¹¹, 2.5:1 (5 women/2 men)²², 3.14:1 (22 women/7 men)⁹, 3.9:1 (263 women/67 men)⁵, 5:1⁶ and 7:1 (7 women/1 men)¹⁰. However, Chandrasekar CR et al¹⁶ described that in a series of 15 elastofibroma cases 12 patients (80%) were men and 3 were women 3 (20%).

ETIOLOGY

The etiology of ED is not fully elucidated. A number of theories have been proposed for its development.

The location of ED between the scapula and the chest wall and the initial description of the tumour in people with hard manual labor constituted the basis for the theory

of mechanical friction between the chest wall and the lower tip of scapula as a mechanism of ED development¹³.

According to this theory, direct mechanical stress on the elastic tissue can cause hypertrophy and secondary degeneration of the elastic fibers, as well as diffuse growth of collagenous tissue³⁰ alternating with the deposition of hyperplastic fat¹³.

However, the higher incidence of ED in women and its location at anatomical sites not involved in mechanical overload can not support the mechanism of mechanical friction, at least as the unique cause, for the development of ED²². In addition, there have been described cases of ED with absence of this causative agent, such as in patients with no history of recurrent or previous trauma in the tumor area or in patients who were not hard workers^{10,22,29}.

Development of ED as a result of the normal age process rather than an abnormal elastogenesis or degeneration, was supported by Geibel et al³¹. This view was based on finding "pre-elastofibroma changes" in their autopsy series. These changes are determined by a weakly eosinophilic material which does not express a definitive elastic tissue formation.

Nagamine et al⁸ supported the familial predisposition for the formation of ED. This view was based on the finding that of 170 cases of ED 32% of cases occurred within a single family. Familial predisposition with an underlying enzymatic defect have also been proposed as possible etiologic factors¹³.

Jarvi OH and Lansimies PH³⁰ argued that in addition to direct mechanical stress on elastic tissue, nutritional deficiency due to failure of the vascular system against friction of the scapula and stretching movements of the upper extremities may play a major role in necrotic tissue changes.

The nature of the abnormal elastic fibers is questionable and controversial and can not substantiate their mechanism of formation. Abnormal elastogenesis or degeneration as a secondary process, even combination of both mechanisms may be responsible²².

Di Vito A et al³³ in a study of extracellular matrix and histopathogenesis in ED, argued that a mechanical strain-dependent reactivation of periostin and tenascin-C expression, as well as elastin deposition, could be responsible for the development of ED.

Kakudo N et al³⁴ in a histochemical, immunohistochemical, and ultrastructural study of an ED, argued that although fibroblasts can produce large amounts of elastin, microinid and collagen, elastin deposition on collagen fibers may be involved in the formation of

abnormal elastic fibers.

Although the mechanism of ED development is not fully elucidated, it is internationally accepted that it is more often in individuals performing repetitive manual labor involving the shoulder^{5,35}, perhaps in combination with genetic factors²⁹.

CLINICAL PRESENTATION

The presence and type of symptoms of ED depend on its location and size²².

ED is usually asymptomatic^{22,29}, especially during the initial phases³⁶.

In these cases ED is accidentally detected by the patient himself, eg in the mirror, or by another person, such as a spouse or member of the family^{7,16,29}, or during physical examination^{5,7}, investigation for other disease¹² or for symptomatic ED on the contralateral side^{5,7,10} or during surgery for unrelated chest injury⁷.

In symptomatic cases, patients usually experience a long history of slowly growing swelling in the shoulder area for which the patient is not looking for medical attention either because the mass is asymptomatic or because the symptoms are mild.

The interval between the detection of swelling or the presence of symptoms until the search for medical assistance has been described in 3-60 months (mean duration of symptoms 20 months)¹⁶, 4 months to 4 years²², seven months¹³ and five years³⁵.

When symptoms are present, these are typical and include local swelling of the shoulder¹⁰, limitation of movement of the upper limb, mainly in the upward movements requiring slipping of the shoulder with respect to the chest wall^{9,35}, and sometimes pain during the movement of the shoulder^{5,16,29,35-37}.

The pain is usually moderate, but cases of painful swelling of the shoulder have been described^{11,35}.

Other symptoms include a clunking sensation during abduction and adduction of the arm^{7,13} and snapping of the scapula²⁹.

PHYSICAL EXAMINATION

The clinical examination reveals a large, palpable, solid mass well circumscribed in the lower corner of the shoulder, usually painless. The mass may be mobile or immobile due to its attachment to the chest wall^{13,16,29,35}.

The tumour is more prominent on the forward flexion

of the shoulder due to forward movement of the inferior angle of the scapula^{16,29,35}.

Small elastofibromas are not visible in the neutral position with the risk of escaping unless the patient is called to move his arm laterally or posteriorly²⁹.

Another mass may be present in the opposite shoulder, often smaller and clinically silent³⁵.

IMAGING FINDINGS

The typical diagnostic pattern of ED is characterized by alternating bundles of fibrous and adipose tissue.

CHEST RADIOGRAPHY

Chest X-ray is usually normal. Except a possible soft tissue signal intensity or elevation of the shoulder, simple X-rays do not show specific changes capable of diagnosing^{22,35,38}.

COMPUTED TOMOGRAPHY (CT) AND MAGNETIC RESONANCE IMAGING (MRI)

EDs have typical imaging findings in CT and MRI (Figures 1, 2a, b) which allow definitive diagnosis^{7,38}.

Typical imaging of ED in CT and MRI include a poorly circumscribed mass of soft tissues without wall. The mass is heterogeneous with a characteristic layered pattern of



FIGURE 1. Chest computed tomography of a 62 years-old woman reveals bilateral solid formations (arrows) with skeletal muscle density located in lateral thoracic walls at the level of the scapulae. (Adapted from Reference¹⁴ after permission).

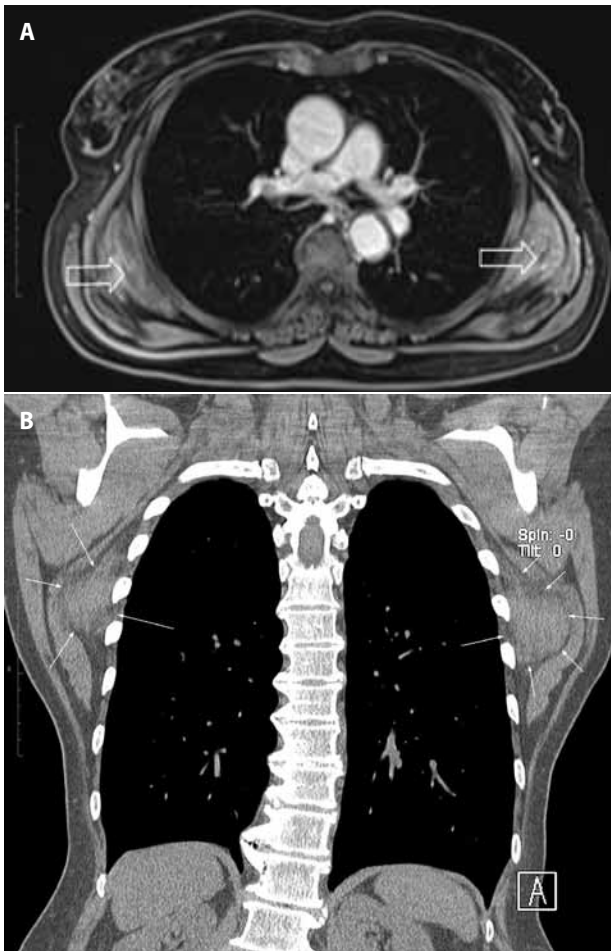


FIGURE 2 (a, b). Chest magnetic resonance imaging with intravenous contrast material of the same patient shows (*arrows*) bilateral unencapsulated symmetrical spindle shaped solid formations located at lateral chest walls at the level of the scapulae, in front of the serratus anterior muscle and lateral to the ribs. The formations are heterogeneous with alternating linear regions of skeletal muscle and fat tissue intensity. Both formations were of similar dimensions, 10×7×3.5 cm. (Adapted from Reference¹⁴ after permission).

fatty tissue (low-density by CT, high-signal on T1 images and intermediate signal on T2 images by MRI) and fibrous tissue (similar to muscle in terms of density by CT and signal intensity by MRI)^{7,38}. The lesions are located anterior to the scapula (subscapular) or caudal to the inferior pole of the scapula (infrascapular), deep in relation to the latissimus dorsi, rhomboid, and serratus anterior muscles. There is no evidence of infiltration of neighboring tissues⁷.

MRI is the imaging modality of choice in diagnosing ED because it depicts better than CT the characteristic

layered pattern. The mass is well-defined. Slight or moderate enhancement within elastofibromas can be observed after gadolinium administration^{5,7,13,35}. However, in some cases of ED, strong enhancement can be observed after gadolinium administration which is characteristic of malignant tumors. In these cases biopsy and histopathological assessment will be required for definitive diagnosis^{35,38}.

In computed tomography the ED displays a typical or a non typical imaging pattern.

The typical appearance of ED in the CT includes a subscapular lenticular mass of soft tissue, with no wall, of equal density to the adjacent muscles (fibrous tissue) and linear areas of low density (fatty tissue). Mass margins show poorer differentiation from surrounding muscles compared to ultrasound and MRI^{7,13,22,35}. This CT pattern is diagnostic for ED.

The atypical appearance of ED in CT includes well-described soft tissue masses with a similar density to the adjacent skeletal muscle, relatively homogenous, with no defined interspersed areas of fat attenuation⁷. This atypical pattern is usually observed in cases of smaller masses³⁶.

The fact that ED presents in the CT and MRI a density/signal intensity similar to that of the adjacent muscles interprets the escape of the EDs from the initial interpreting radiologists. Naylor MF et al⁷ described that of 21 fibroblastomas imaged, only four (19%) were perceived by the initial interpreting radiologist, and only one (5%) was correctly diagnosed⁷. Brandser EA¹² described that none of 5 fibroblastomas in 4 asymptomatic patients had been described in the initial diagnosis of CT.

PET-CT SCAN

In PET-CT scan most tumors appear with mild or moderate diffuse metabolic activity with the SUVmax ranging from 1.4 to 3.2⁹ which should not be interpreted as a malignant finding³⁶. However, hypermetabolic tumors have been described³⁹.

ULTRASOUND

EDs present in the ultrasound a typical pattern of a well-defined soft tissue mass in the typical position of the ED, with a multilayered pattern of alternating layers of hypoechoic fat and hyperechoic elastic tissue, gently parallel to the chest wall⁴⁰⁻⁴². Colour Doppler shows a vascular pattern similar to the surrounding muscles²².

Although ultrasound represents an inexpensive di-

agnostic tool²², it is not an imaging technique of choice for diagnosing of ED. In addition, the examination is dependent on the examiner and for this reason it is not always diagnostic³⁵.

DIAGNOSIS

The diagnosis of ED can be definitively determined on the basis of its clinical and imaging characteristics.

A history of long-term presence of a solid mass under the scapula⁴³, the bilateral location⁷ and the classical imaging characteristics described above in elderly patients and especially in women place the diagnosis of ED. In these cases, the biopsy is not necessary for diagnosis^{5,13,22,16,29,35}.

Biopsy is necessary to confirm diagnosis in cases of unilateral EDs without the typical imaging pattern in MRI and CT scan^{5,7}.

Particularly unilateral mass of soft tissues larger than 5 cm in diameter in elderly patients, with MRI images of weakly defined, heterogeneous mass of soft tissue, sometimes enhanced by gadolinium, should be identified by biopsy because of the possibility of soft tissue sarcoma in this age group^{16,44}.

Fine needle aspiration biopsy (FNAB) is a commonly used technique to definitively diagnose the condition^{10,13}. However, the collagenous nature of the mass often results in a paucicellular FNA smear, resulting in a false-negative report^{5,10,22}. In these cases, a core or open biopsy is essential to get a representative tissue specimen to make a definitive diagnosis^{5,22} especially in case of suspicion of a soft tissue sarcoma¹⁵.

The differential diagnosis of ED includes sarcoma, lipoma, fibroma, liposarcoma fibromuscular tumour, desmoid tumour, hemangioma, hematoma and aggressive fibromatosis^{5,35}.

PATHOLOGY

Macroscopically, the fibrolastoma is characterized by a solid, slightly elastic, irregular, weakly defined mass of fat and connective tissue without wall with a consistence that assimilates rubber. The cut-surface shows white and yellow tissue caused by fat trapping, similar to a check-board pattern²².

Microscopically, the fibrolastoma is characterized by the presence of a large number of abnormal elastic fibers embedded in a matrix of dense eosinophilic collagen, benign fibroblasts, and mature adipocytes^{7,16,22}.

Elastic fibers are difficult to be detected by hematoxylin-eosin staining²². They are best depicted by elastin staining^{7,22}, which reveals deep-stained branched and unbranched fibers⁷. The elastic fibers are sometimes elongated and larger than regular ones, mostly round shaped, densely packed²², simulating beads on the strings¹³.

The fibroelastic fibers exhibit a central dense core and serrated margins^{7,45,46}. The central core represents a mature elastic tissue that appears to be secreted by active fibroblasts⁴⁶. The peripheral zones of elastic fibers consist of a variety of fuzzy, irregularly shaped amorphous components and compactly and randomly arranged large quantities of microfibrils⁴⁵.

The lesions are predominantly subcellular with fibroblastic cells without atypia and mitotic activity²². In the cytoplasm of the fibroblasts there are dense granular bodies, which are believed to represent elastin or elastin precursors⁴⁶. Between the benign fibroblasts there is a varied amount of fatty tissue^{7,16}.

MANAGEMENT

When diagnosis of ED is definitive, its treatment depends on the symptoms it causes to the patient.

In asymptomatic patients, clinical monitoring is adequate⁵ due to the benign nature of the tumor and because malignant transformation has never been described^{13,22,25}.

Surgical excision is indicated when there is a diagnostic doubt¹⁰ and in severe symptomatic cases of snapping or blocking scapula, pain, or tumor-related discomfort and depending on the patient's psychological and physical strain. A therapeutic marginal excision has been shown to be sufficient and it is preferred over broad or radical excision^{5,10,22,29,35,44}.

The most frequent complication observed postoperatively is hematoma due to the fact that the periscapular region is highly vascular^{13,29}.

OUTCOME

The prognosis after surgical excision is good^{13,37}.

Tumor relapses have been described in 7% of the cases and are attributed to incomplete resection of the tumor¹⁰.

CONCLUSIONS

ED is an unusual, well defined tumor of soft tissue typically located in the peri-scapular region, affecting mainly

elderly people and especially women. The typical location and the characteristic MRI and CT imaging pattern of the alternate bundles of fibrous and adipose tissue set the diagnosis without the need for biopsy. Biopsy is required in atypical cases to exclude malignancy, especially sarcoma. In asymptomatic cases, simple monitoring is sufficient. Surgical excision is recommended for serious symptoms and diagnostic doubt. The prognosis is excellent. No case of malignant transformation has been described.

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