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Personal experience and a review of the literature



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Surgical treatment of elastofibroma dorsi. Personal experience and a review of the literature

AIM: To report our experience in treating elastofibroma, an uncommon lesion, usually arising into subscapular area; it has been included between soft tissue tumors and is characterized by progressive growth and benign behavior

METHODS: Patients with an histologically proved elastofibroma, operated at our ward unit over a 3-year period, entered this study. Early results of surgical treatment have been analyzed and compared to those of Literature, focusing on selection criteria, hospital morbidity and relative risk factors

RESULTS: Fourteen surgical procedures have been performed on 11 patients; EF presented as bilateral on 3 patients (27.3%) and these patients were treated with sequential 2-stages excision. All patients received complete surgical resection according to marginal excision technique; mean operative time was 75.8 ± 21.4 min. (range 55-135) while mean size of resected EF was 8.57 ± 2.2 cm. (range 5-12). Three patients developed significant postoperative seroma (21.4%), while neither hemorrhages nor recurrences have been observed. Increased B.M.I. was the only factor significantly related to hospital morbidity at univariate analysis ($p = 0.0339$)

CONCLUSIONS: Patients carrying elastofibroma larger than 5 cm. and symptomatic should undergo surgical treatment; marginal excision represents the standard technique; we recommend the use of ultrasound energy device for tissue dissection: its current use seems to prevent postoperative bleeding. Development of postoperative seroma seems related to increased patient's B.M.I. and to larger size of EF, rather than to different methods of dissection.

KEY WORDS: Chest wall tumors, Elastofibroma dorsi, Elastin, Marginal resection, Soft-tissue tumors

Introduction

Elastofibroma Dorsi (EF) was first reported by Jarvi and Saxen in 1961 as a benign lesion arising into subscapular region and characterized by a slow growth and by the proliferation of elastin fibers into a stroma of collagen and fatty tissue ¹. In 2002 the World Health

Organization defined EF as a benign fibroblastic/myofibroblastic tumor and classified it among soft-tissue tumors ². Since then, less than 600 cases have been reported in Literature, mainly from case reports and small consecutive series; to date, few consistent clinico-pathological analysis may be considered and compared for their results ³⁻⁶. Large reviews from oncologic database and soft-tissue tumor register, show a prevalence for EF from 0.086% to 1% among all soft-tissue tumors, while incidence rises to 1.5-15% considering radiological or autopsy series ⁷.

EF is usually located in the soft tissues of the inferior angles of the scapulae, deep between latissimus dorsi, serratus anterior and rhomboid muscles, and lateral to the periosteum of the 6th-8th ribs and intercostal muscles. Occasional sites (less than 5%) include hands, feet,

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mouth, joints, mediastinum and aorta⁸; abdominal elastic deposits, such as fibroelastosis, angioelastosis and EF have also been reported arising from mesentery, greater omentum, spleen and peripancreatic fat^{9,10}.

EF usually occurs between the fourth and the seventh decade of life and is more common on female (ratio Male to Female 1: 2/2.5)¹¹; it has been described as bilateral in a rate ranging from 15% to 60%¹², while no significant differences have been reported between right-sided and left-sided lesions⁶.

Patient usually complains of local pain and functional limitation of the shoulder while physical examination shows distinct subscapular palpable lump, more evident with abduction and elevation of the arm. Lesion may also completely disappears beneath scapular area with shoulder movements and scapular snapping is also described. Clinical examination and imaging assessment lead to the diagnosis of EF with a high degree of confidence: ultrasonography (US), computed tomography (CT) and magnetic resonance imaging (MRI) have been proposed to assess diagnosis, but they are adopted in different combination in the clinical practice, as no univocal diagnostic algorithm has been reported in the literature¹³⁻¹⁵.

Occasional experience with Positron Emission Tomography-CT (PET-CT) has been reported¹⁶. Even if any technique can be defined as the gold standard, presence of reliable imaging tools and physical findings make preoperative cytology as unnecessary¹⁷.

Pathogenesis of EF is still a source of debate: several mechanisms have been proposed but to date any definitive conclusions can be stated. Developing of EF is frequently associated to manual and heavy shoulder activities: a rate ranging from 30% to 60% of patients reports of repeated mechanical shoulder movements, as a consequence of their job, sportive activity, hobbies or housework; repeated microtraumas due to the friction of the tip of the scapula to the thoracic wall may cause inflammation and degeneration of the elastic fibers¹⁸.

According to this mechanical theory, hyperproliferation of the fibroelastic component can be related to tumor (or pseudotumor indeed) development. On the other hand, a certain genetic component has been hypothesized: approximately 30% of patients carrying EF exhibits some form of familiar predisposition. In these patients, resected specimens often show some genetic anomalies, such as DNA sequences losses, chromosomal instability and/or presence of transforming growth factor β and basic fibroblast factor¹⁹. Following this theory of genetic predisposition, EF could be considered as a real neoplastic process rather than a result of repeated microtraumas⁶. Finally some authors argued that EF could represent a simple consequence of aging: vascular insufficiency and suffering of periscapular tissues may result in hyperproliferation of fibroblasts and hyperproduction of elastic fibers¹⁸.

Surgery is the only treatment for EF; it is usually indi-

cated for symptomatic patients (significant pain, increasing swelling and/or shoulder impaired function) and for larger lesions: 5 cm. in maximum size is the accepted cut off value^{20,21}. Further indication may result from unusual clinical and/or radiological findings, which may suggest differential diagnosis with soft tissue sarcoma, desmoid, neurofibroma or subacute hematoma²². In these unusual cases also preoperative cytology or biopsy may be considered. Surgical technique usually deals with marginal excision, which consisted in the dissection of peritumoral tissues, using electrocautery or ultrasonic device. A surgical margin of 5 mm is considered as adequate for this type of resection^{5,6,23}. Tumoral bed is usually reached by muscle-sparing technique and both latissimus dorsi and serratus anterior muscles are dissected along their fibers and splitted; EF appears as a un-capsulated, whitish, hard and ill-defined mass, fixed to the scapular periosteum or to the external thoracic fascia. Neoplasm has to be completely removed (R0 resection) preserving muscle layers³; sometimes partial detachment of the rhomboids have been reported⁴. Histological examination confirms diagnosis of EF as well as the complete excision of the lesion and absence of residual tumor. Due to patient's lateral decubitus on operative setting and in order to decrease postoperative sequelae, bilateral EF are usually treated with sequential two-stage excision. Removal of larger lesion is followed by a second procedure after an interval of 12-24 months²⁴.

EF still remains an uncommon soft-tissue neoplasm and some aspects of its clinical behavior still need to be better defined; as we collected a consistent cohort of patients operated for EF in a 3-years period, herein we report our results, compare them to those of other consistent series and discuss main features of this rare entity.

Methods

Patients operated from May 2018 to May 2021 at our ward unit and with an histopathologically confirmed EF entered the study and were retrospectively reviewed. Data were collected by reviewing patient's medical records, surgical register, pathological response and through follow-up dial interviews. Patients were asked in particular for their jobs, hobbies, houseworks or any heavy manual activities as well as for correspondence between lesion side and dominant hand.

All patients were addressed to MRI, while 7 patients (63%) and 5 patients (45%) have just received US and CT respectively at the time of first evaluation. We considered MRI as mandatory because of its highest ability in differentiating EF from other soft tissue tumors. All cases presented EF larger than 5 cm. in maximum diameter. Functional results were assessed by using Constant-Murley score, which considers 4 variables: pain, ability in daily activities, Range of Motion (ROM) and

strength of abduction²⁵; evaluations have been made and compared at the time of first outpatient approach and after 60 days since surgical procedure.

Patients had just been operated under general anesthesia and all procedures had been performed by the same senior general surgeon with similar technique (GL). Patients with bilateral EF underwent sequential two-stages excision, having firstly operated larger and more symptomatic EF.

On operative setting, patient was positioned in a contralateral decubitus, with the ipsilateral arm extended anteriorly, in order to allow the lump to slight downward the angle of the scapula and to obtain an optimal exposure of the tumor. Scapular margins and tumor boundaries were marked preoperatively (Fig. 1).

An oblique 10 to 12 cm skin access was performed over the lesion and below the tip of the scapula. A muscle-sparing technique has always been adopted both for latissimus dorsi and serratus anterior muscle, so fibers were dissected along their course. EF usually appears as a translucent, nacreous and white to grey defined mass, strongly attached to the periosteum of the ribs and the muscular fibers. We always used an ultrasound shear device in order to obtain accurate marginal excision and to minimize postoperative blood loss and serum effusion;

it provides a safe dissection also in a very narrow space, like subscapular area. After excision, a tubular section drainage was placed and the integrity of muscular layers was restored with interrupted absorbable sutures. A compression bandage was applied and maintained for 7-10 days. No particular restriction to shoulder movements was adopted in the postoperative period. Drain was kept in place for at least 24 hours; in case of prolonged drainage time, the patient was discharged with the drain in place. All patients received postoperative medications and controls at our outpatient service. Resected specimens typically appear as a bulky pear-shaped lesions, with a root arising beneath scapulae and rhomboids (Fig. 2); all specimens were addressed for definitive histological study both with standard hematoxylin-eosin stain and with elastin histochemical stain (Fig. 3).

Patient's data were collected into an Excel 2016 Spreadsheet (Microsoft Office®); statistical analyses has been performed using the SPSS® statistical software 25.0 (SPSS Inc, Chicago, IL, USA). Descriptive analysis is expressed in terms of frequency, mean \pm SD, median and range. All variables were matched to each other in a univariate analysis; Chi-square test and the Fisher exact test were used to compare variables. Statistical significance was defined as $p < 0.05$.



Fig. 1: EF: patient's positioning on two operative settings. Scapular margins and tumor boundaries are marked. Evidence for contralateral smaller EF (arrow).

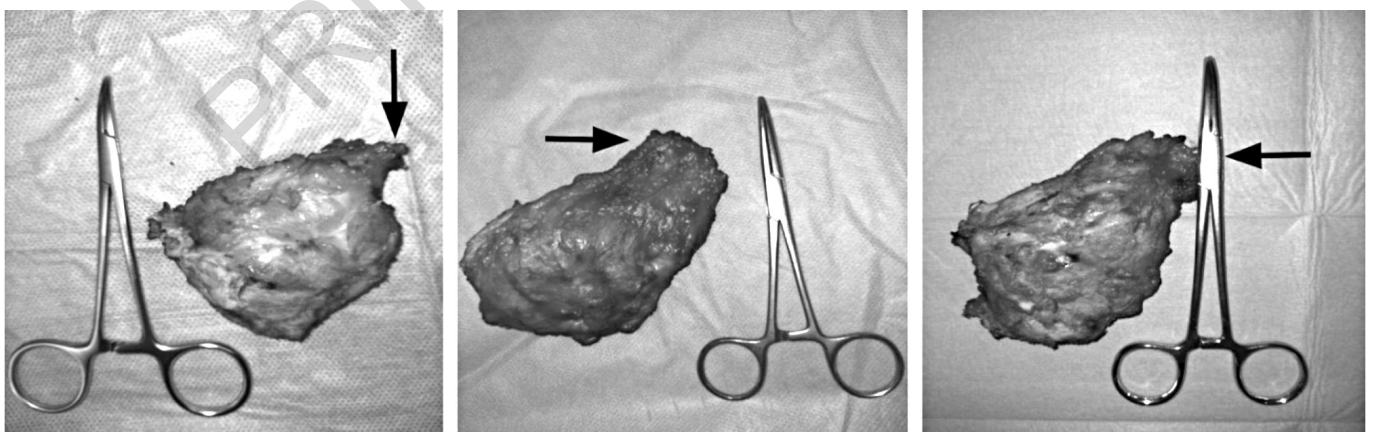


Fig. 2: EF: macroscopical appearance on 3 resected specimens. Root of EF arises beneath scapulae and rhomboids (arrows).

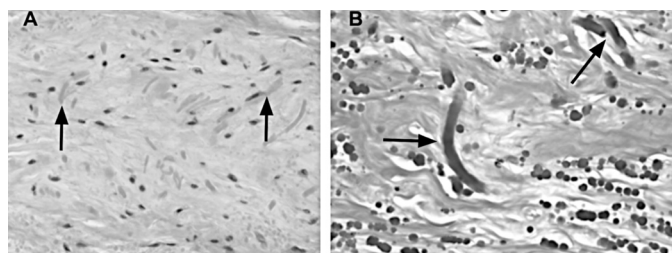


Fig. 3: EF: Microscopic findings; (A) alternance of elastic connective and fibrous tissue (Hematoxylin & Eosin stain 20x; arrows indicating elastic fibers); (B) coarse elastic fibers appear arranged in irregular shape (arrows) (Histochemical stain for elastic fibers 40x).

Results

Fourteen surgical procedures have been performed on 11 patients; EF presented as bilateral on 3 patients (27.3%) and both lesions have been just detected at the time of first diagnosis. All EF were located at the angle of the scapula.

Patients were 6 females and 5 males, with average age at the time of diagnosis of 62.4 ± 9.2 years (range 49-80). No significant side prevalence was detected: 5 patients exhibited left-sided EF (62.5%) and 3 patients right-sided EF (37.5%). Length of disease complaints ranged from 18 to 65 months, while a significant history of heavy manual activity from job, hobbies or housework could be demonstrated on 4 patients (36.4%).

All patients referred symptoms like pain, increased swelling and significative tissue enlargement when examined at our outpatient service, with increasing limitation in their upper limb movements. Correspondence between lesion side and dominant arm was reported on 5 patients (62.5%).

The mean BMI of resected patients at the time of diagnosis was 26.7 ± 2.5 (range 21.8 - 31.1), while 2 patients presented a long history of antiplatelet oral therapy (18%). Those patients carrying bilateral EF had second operation after 11, 16 and 18 months since first surgical procedure. Patients were classified as ASA II in 10 cases (91%) and ASA III in 1 case (9%) at preoperative aenesthesiologic work up.

All patients received complete surgical resection according to marginal excision technique; mean operative time was 75.8 ± 21.4 min (range 55-135) while mean size of resected EF was 8.57 ± 2.2 cm (range 5-12). Wound drainage with negative pressure was always left in place at the end of operation and 4 patients have been discharged with drainage because of prolonged serum effusion (28.5%). Mean length of hospital stay was 2.5 ± 0.7 days (range 2-5) while mean drainage time was 3 ± 1.3 days (range 2-6).

Three patients developed significant wound seroma, which required repeated needle aspiration at our outpatient service (21.4%), while no wound hematoma or postoperative haemorrhage have been observed. Thanks to muscle-sparing surgical technique, patients did not develop significant shoulder impairment and none required postoperative rehabilitation.

Comparison between pre- and postoperative Constant-Murley score showed a significant improvement in shoulder function (64.31 ± 7.02 and 89.08 ± 6.78 respectively, $p < 0.05$). Mean length of follow up is 14.7 ± 8.1 months (range 4 - 33) and to date no recurrences have been observed.

Increased BMI was the only factor significantly related to postoperative morbidity at univariate analysis ($p = 0.0339$); also EF mean size appeared related to increased morbidity even if difference did not reach statistical significance ($p = 0.0894$) (Table I).

Discussion

EF is a rare soft-tissue benign tumor, usually arising in subscapular area; surgical resection represents the main treatment, even if some results have been reported with radiotherapy²⁶. Criteria for surgical selection are still unclear: age of patient, presence of heavy shoulder impairment, size of the lesion and possibility for a different diagnosis, such as sarcoma or metastases must be always taken into account; usually patients with lesion larger than 5 cm and with a significant impairment in their ROM may be addressed to surgery (23). We followed these selection criteria, considering that preoperative imaging often underestimates real size of EF: El-

TABLE I - *Elastofibroma dorsi (EF): prediction of morbidity univariate analysis*

	Morbidity +	Morbidity -	Statistical Analysis		<i>p</i>
Mean Age (yr)	62.33	62.37	T test	-0.005243	0.9959
Gender (F/M)	1/2	5/3	O.R.	0.3	0.545
B.M.I.	28.5	26.03	T test	-2.455448	0.0339
Mean EF Size (cm.)	11.5	8.83	T test	-1.88108	0.0897
Operative Time (min.)	87	83.6	T test	+0.199276	0.846
Anti PLT Therapy	1/2	1/7	O.R.	0.2857	0.99
			C.I.95%	0.0118-6.9142	

TABLE II - Elastofibroma dorsi (EF): literature review - Patient's demographics and results on consecutive series

Author Year	F. Lococo ¹ 2013	El Hammouni ² 2014	M. Ali Deveci ³ 2017	A. Scamporrino ⁴ 2020
N° Patients	71	76	51	59
Age	60.2 ± 8.8	49 (38-70)	55.3±7.01	59 (33-81)
Gender F/M	48/23	54/22	45/6	35/24
Bilateral	12/71 (16.9%)	20/76 (26%)	19/51 (18.6%)	11/59 (18.6%)
Heavy Activities	42/68 (61.8%)	19 (25%)	5 (9.8%)	27 (46%)
Size	7.2 ± 2.1	6-18 cm	8.7 cm (5-13)	7.94 ± 1.89
Length Hosp. stay (days)	3 ± 1.2	n.r.	n.r.	2 (1-9)
Hosp. Morbidity	10.6%	9 (11.8%)	11.5%	12 (17%)

1. Thorac Cardiovasc Surg 2013; 61 (3): 215-222

2. Korean J Thorac Cardiovasc Surg 2014; 47(2): I I 1-116

3. Acta Orthop Traumatol Turc 2017; 51(I): 7-1 I

4. J Thorac Dis 2020; 12(5): 1884 - 1894

Hammouni et al report a clinical/surgical diameters ratio of 0.53 in their large experience ⁴.

Surgical resection by using marginal excision is the currently accepted technique; some alternative surgical procedures for EF include latissimus dorsi muscle flap; they have been proposed by aesthetic surgeons, but were found related to higher length of procedure and significant seroma incidence in the donor site ²⁷; combination of quilting sutures and fibrin sealant may reduce the incidence of postoperative seroma at the donor site ²⁸. Also resection with scapula-thoracic joint arthroscopy has been described for selected cases ²⁹.

Concerning early results of surgery, we examined four large consecutive series, 3 from Thoracic Units ³⁻⁵ and one from Orthopedic and Traumatology Unit ⁶, dealing with more than 50 patients operated and with an histological definitive diagnosis of EF (Table II). In these studies, data have been collected by retrospective cohort analysis or observational information from patient medical records, surgical records, pathological examination results.

Early follow-up has been always obtained by phone interviews or outpatient clinical visits; after that open-door policy has been usually adopted. Study period ranges from 5 years ⁴ to 10 years or more ^{3,5,6}. All series report a female prevalence, while rate of bilateral EF ranges from 16.9% to 26% and no clear relationship with heavy manual activities can be demonstrated (rate from 9.8% to 61.8%). Hospital morbidity ranges from 10.6% to 17% of patients and the most common complications result wound effusion with seroma and postoperative hematoma; less frequent complications include wound infection, skin ulceration and parietal textiloma ^{4,30}. Some patients are reported requiring reoperation for drainage of hematoma and wound debridement ^{3,6}.

Development of wound effusion is usually related to the size of the lesion or insufficient fixation: in order to prevent this complication wound drainage with negative pressure, bandage compression and postoperative limb

immobilization are recommended. Larger tumor size ($p = 0.005$) and assumption of antiplatelet medications ($p = 0.038$) were related to increased morbidity at univariate analysis ^{3,5}, while a BMI < 25 was the only independent factor related to decreased morbidity at multivariate analysis ($p = 0.024$) ⁵. In our experience, we always used surgical drain and bandage compression in order to minimize morbidity, while avoided prolonged shoulder immobilization and encouraged patients to gradually resume their activities; drainage has been removed during postoperative stay or at outpatient control according to the amount of discharge. No reoperation have been required. Significant rate of postoperative seroma is also confirmed in other series ^{21, 31-33}. Postoperative hematoma is less frequent but may require reoperation and longer postoperative stay: Nagano et al. reports an incidence of 9 postoperative hematomas on 20 EF operated (43%); they have been treated with conservative treatment on 8 cases, while one patient required surgical evacuation. Factors affecting the development of postoperative hematoma resulted larger tumor size ($p = 0.02$) while duration of postoperative drainage was significantly longer in non-hematoma group ($p = 0.001$). This high incidence of hematoma discourages surgical treatment of EF in asymptomatic cases ²³. Postoperative hematoma after surgery for EF has been reported as high as 38.9-87.5% in some series ^{17,22,33}; in order to reduce this complication, Nagano et al recommend a week of limb immobilization after surgical procedure and duration of wound drainage for at least one week. Ali Deveci et al reported an 11.5 % morbidity rate on 61 procedures performed on 51 patients (4 seromas and 3 postoperative hematomas). One patient required reoperation for hematoma drainage. He found no correlation between tumor size and postoperative morbidity ($p > 0.05$) ⁶. Lococo et al recommend meticulous hemostasis during surgery for EF; they report a 19.7% morbidity after EF resection ¹⁴⁻⁷¹. In their experience drainage may be removed during hospital stay or after

discharge at outpatient clinic, according to progressive reduction of output. They report one case of postoperative bleeding which required revision of hemostasis for a very large EF (14 x 10 x 7 cm)³. Few authors emphasize method of dissection as possible variable for postoperative bleeding complications: marginal excision is usually performed by using smooth finger dissection, scissors or electrocautery⁵. In our experience we recommend the use of ultrasound harmonic device: it is very useful for accurate peritumoral dissection and delivers precise energy to limit lateral thermal spread and to protect critical structures during dissection. Furthermore it maximizes visualization and control in a very narrow space, like subscapular area.

EF is a benign tumor and local recurrences should be related to unproper surgical technique rather than to its biological behavior. Some relapses have been reported whenever patients referred to hub hospital coming from peripheral centers³. No recurrences have been reported on other large series after consistent follow up⁴⁻⁶.

Conclusions

In our experience patients bearer of EF larger than 5 cm and symptomatic should undergo surgical treatment; MRI represents the gold standard imaging technique, because of its ability in differentiating EF from other soft tissue lesions. We didn't find significant correlation between EF and manual activities and always treated bilateral lesions in two different stages. Surgery has been performed by using marginal excision technique; we recommend the use of ultrasound energy device for tissue dissection: its current use seems to prevent postoperative bleeding. Development of postoperative seroma seems related to increased patient's BMI and to larger size of EF, rather than to different methods of dissection.

Riassunto

SCOPO: Descrivere i principali aspetti clinici dell'elastofibroma, una rara lesione generalmente localizzata in corrispondenza della regione sottoscapolare; viene classificata fra i tumori dei tessuti molli ed è caratterizzata da un accrescimento progressivo e da un comportamento biologico benigno. Meno di 600 casi sono stati descritti ad oggi nella Letteratura Internazionale.

METODI: I pazienti operati nel periodo Maggio 2018 - Maggio 2021, con una diagnosi istologica definitiva di elastofibroma, sono oggetto della presente descrizione. I risultati a breve termine del trattamento chirurgico sono stati analizzati e comparati con quelli della Letteratura recente, con particolare riferimento ai criteri di selezione per il trattamento chirurgico, alla morbilità ospedaliera ed ai fattori di rischio per la morbilità stessa.

RISULTATI: Sono state eseguite 14 asportazioni su 11 pazi-

enti; la lesione era bilaterale in 3 casi (27,3%), e questi pazienti sono stati trattati con due exeresi in tempi separati. Sono state effettuate exeresi radicali, in accordo con la tecnica della escissione marginale; il tempo chirurgico medio è stato di 75,8 ± 21,4 min. (range 55 - 135), mentre il diametro medio delle lesioni è risultato 8,57 ± 2,2 cm (range 5 - 12). Tre pazienti hanno sviluppato un sieroma postoperatorio clinicamente evidente (21,4%), mentre non sono state osservate complicanze emorragiche né recidive durante il follow up. L'analisi univariata dei fattori di rischio per sviluppo di complicanze ha dimostrato l'incremento del BMI come unico fattore statisticamente correlato ($p = 0,0339$).

CONCLUSIONI: Pazienti con elastofibroma superiore ai 5 cm di diametro e sintomatici devono essere sottoposti ad exeresi chirurgica, con la tecnica della escissione marginale. Consigliamo l'utilizzo del dissectore ad ultrasuoni per la manipolazione tessutale: il suo uso routinario sembra prevenire le complicanze emorragiche PO. Lo sviluppo di sieroma sembra invece correlato al BMI del paziente ed alle dimensioni della lesione, più che alle differenti metodiche di dissezione.

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