

Case Report

Giant malignant Chondroid Syringoma: Case Report and Literature Review

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Abstract

Chondroitin syringoma, also known as cutaneous mixed tumor, is a rare type of sweat gland tumor, accounting for 0.01% to 0.1% of all primary tumors of the skin. The malignant form is extremely rare, with 41 cases described so far. It predominates in the trunk and distal extremities and affects women more frequently, with a 3:2 rate 6,7. Patients may reach the health service with large lesions due to the fact of being slow-growing lesions and have a high rate of recurrence, requiring surgical treatment that may lead to extensive defects making complex reconstructions necessary. We present a case of malignant chondroid syringoma in an 80 years old male patient, with 29 years of evolution and unusual size. A reconstruction was performed with a muscular dorsal flap of the latissimus dorsi with good aesthetic and functional results, with no signs of recurrence after 8 months of follow-up. It is concluded that because it is a late diagnosis neoplasia, the surgical treatment ends up generating expressive defects. There for musculocutaneous should be considered as an option for reconstruction, aiming optimal functional and aesthetic restoration.

Keywords: Latissimus dorsi flap; Malignant chondroid syringoma; Mixed skin tumor; Upper limb reconstruction

Introduction

Chondroid syringoma, also known as cutaneous mixed tumor, is a rare type of sweat gland tumor, accounting for 0.01% to 0.1% of all primary tumors of the skin [1,2]. It is a benign lesion, histological and immunohistochemical similar to the benign mixed tumors of the salivary gland (pleomorphic adenoma) [3].

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Most of chondroid syringomas are located in the head and neck region (80%), commonly involving the nose and malar region, and reach dimensions smaller than 3cm, although tumors up to 10cm have already been described. They occur normally in the sixth decade of life, predominantly in men, with a ratio of 2: 1 [1,4].

The malignant form is extremely rare, predominant in the trunk and distal extremities, presenting as a firm, subcutaneous nodule of slow growth [5], it affects women more frequently, with a ratio of 3: 2 [1,6] and has only about 40 cases described until now [4,6-8].

Diagnosis is made essentially by histological study, and most tumors have a high recurrence rate. It is also worth mentioning that overlapping areas of benign and malignant tumor may occur in a primarily benign tumor [9].

It frequently presents local and / or regional metastases (about 60%), especially for lymph nodes, lungs and bones; In addition, it has a mortality rate of approximately 25% after a long evolutionary course.

First-line treatment is based on the surgical resection of the lesion after performing adequate staging based on imaging studies [1,6,7].

Because these tumors are rare, recurrent and insidious, these patients may reach the health service with lesion of large dimensions. This fact associated with the surgical treatment of excision may lead to the development of extensive defects requiring complex reconstruction [1,10,11]. The involvement of upper limbs is infrequent, but large tumors on this topography requires a type of reconstruction that may be challenging for the plastic surgeon [10]. Thus, the use of the dorsal musculocutaneous flap may be one of the best options to be used [10,12].

We present a case of malignant chondroid syringoma in an 80 years old male patient, with 29 years of evolution and unusual increased dimensions.

Case Report

Male patient, 80 years old, with complaint of right arm lesion with progressive growth lasting for more than 20 years. He presents a surgical history of 6 procedures for tumor's excision, with recurrence in all episodes, and last resection was performed 7 years earlier. Physical exam showed, in the right deltoid region, lobulated tumor of approximately 20 cm, not adhered to deep planes, with central ulceration of approximately 3 cm. Brachial, radial and ulnar pulses were present and without signs of nerve compression (Figure 1). Magnetic resonance imaging of the right upper limb revealed a massive multiloculate formation of approximately 20 x 10 cm, without neurovascular bundle infiltration. Regarding muscular planes invasion, a well defined cleavage plane between the mass and the triceps' belly in the middle and lower thirds of the mass. On the middle third of the arm there was no clear cleavage plane between the mass and the triceps as well as between the anterior component of the mass and the biceps (Figure 2). Other staging exams (chest, abdomen, and pelvis tomography) showed no signs of systemic involvement. An incisional biopsy with diagnosis of chondroid syringoma was performed [13-17].



Figure 1: Magnetic resonance imaging of the right upper limb revealed a massive multilobulate formation of approximately 20 x 10 cm, without neurovascular bundle infiltration.



Figure 2: Anterior component of the mass and the biceps.

Local resection of the lesion and immediate reconstruction of the right upper limb with pedicled latissimus dorsi flap associated with split-thickness skin grafting (Figures 3 and 4) was performed. The anatomopathological result was of malignant chondroid syringoma of 26cm, ulcerated, with areas of necrosis and hemorrhage and areas with capsule and adipose tissue invasion. No angiolymphatic and perineural invasion were detected. All margins were free of disease [18-23].

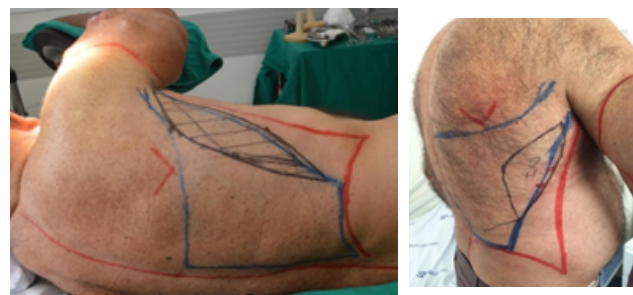


Figure 3: Right upper limb with pedicled latissimus dorsi flap.

The patient progressed satisfactorily, leaving the hospital on the 5th postoperative day. He is currently undergoing outpatient follow-up (Figure 5), with no signs of tumor recurrence after 2 years of surgery.

Discussion

The term chondroid syringoma was first used by Hirsch and Helwing in 1961 to designate a skin tumor previously known as a mixed skin tumor originating from sweat glands. Malignant Chondroid Syringoma is an extremely rare cutaneous tumor, unlike its benign form which is relatively common. According to the MEDLINE database, only 41 cases of malignant form have been reported previously since 1961 [4].

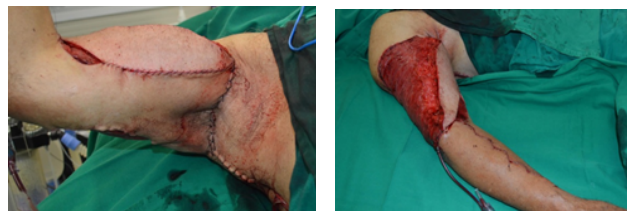


Figure 4: Split-thickness skin grafting.

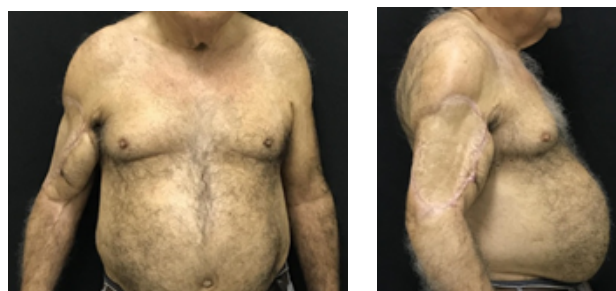


Figure 5: With no signs of tumor recurrence after 2 years of surgery.

Clinically there is no distinction in appearance between the benign and malignant form. Malignant form usually presents as a non-ulcerated nodule. This type of tumor sometimes appears well circumscribed, with cystic appearance, however in other cases it may be adhered to the deep planes.

From 41 cases of malignant chondroid syringoma described so far, 26 occurred in women, and 15 cases in men and with a ratio of 2: 7 in the benign form. The mean age in males was 49.2 years (ranging from 9 months to 70 years of age) (ie distribution by all age groups) and the most commonly affected sites were hands and feet (20 patients), followed By the head (n = 13) and the trunk (n = 8) (Table 1).

The reported case is distinguished by the location in the proximal region of the upper limb and by the large dimensions, with extremely slow evolution in 20 years.

Malignant chondroid syringoma tend to follow an unpredictable clinical course. the reported cases, including ours, more than 34% had local recurrence. It frequently presents local and / or regional metastases (about 63%), with a tendency to invasion of adjacent structures, with cases of bone and CNS invasion. Distant metastases were mainly for lymph nodes, lungs and bones. The mortality rate is approximately 17% after a prolonged evolutive course due to disseminated metastases [9].

	Author	Journal	Year	Age	Gender	location	size (cm)	Local recurrence	Metastasis	Treatment	Evolution
1	Casteleiro Roca, et al. [24]	CIRUGÍA PLÁSTICA IBERO-LATINOAMERICANA	2009	68	F	Left arm	21	no	Left lung 6mm / left axillary lymph node	Extirpation with broad margins and primary closure + chemotherapy (6 cycles with Ifosmamide, Mesna, Adriamycin and Dacarbacin).	DFS 2 years
2	Daniel Chang et al [25]	J Bras Patol Med	2007	54	M	Sternal	0,9	no	no	Extirpation with broad margins and primary closure	DFS 3 years
3	Rajesh Malik, et al. [26]	Indian Dermatol Online J.	2013	61	F	scalp	Great vegetation - pedicle of 8.0	no	Local invasion until dura-mater	Extirpation with margins and primary closure	Death on 2nd PO - meningitis was the most likely cause
4	Arvind Krishnamurthy, et al. [27]	Indian J Nucl Med	2015	41	M	Left ear	3	no	Left cervical lymph node - simultaneous diagnosis	Excision with margins + local flap + left neck dissection	Not available
5	Deniz Tural, et al. [28]	Case Reports in Oncological Medicine	2013	34	F	face	1,5	no	no	Excision with 1,0 cm margins (closure not informed)	DFS 3 year
6	P Shashikala, et al. [29]	Indian J DermatolVenerolLeprol	2004	32	F	scalp	5	no	no	Excision with margins	Loss of follow-up
7	Akira Watarai, et al. [30]	Dermatology Online Journal	2011	46	M	Right foot	3	no	Right Inguinal lymph node 12 years after primary tumor	Excision with margins (t primary tumor) + lymphadenectomy + RT and chemotherapy (metastasis) tegafur, gimeracil, and oteracil potassium	DFS 18 months after metastasis treatment
8	Eiji Ishimura, et al. [31]	Cáncer	1983	60	M		7	yes after 3 years (10 x 4cm)	7 years bilateral cervical lymph nodes / 11 years widespread	Tumor excision, local recurrence and cervical lymph nodes	Death after 11 years
9	Clara Redono, et al. [32]	Cáncer	1982	61	F	Right foot	2,5	yes - 4 previous relapses	Inguinal 7cm, multiple pulmonary nodules,	Chemotherapy vinblastine, vincristine, and Genoxal no response	Palliative treatment and loss of follow-up
10	Vivek Agrawal, et al. [33]	The Journal of Dermatology	1998	40	F	scalp	6	Yes - 3 previous relapses	Bilateral occipital lymph node	Excision with margins + local flap + excision of affected lymph nodes + RT post-op	DFS 25 months
11	S Nicolaou, et al. [34]	Australasian Radiology	2001	54	M	Right hand	4,5	no	no	Excision with margins	not informed
12	Joaõ Luiz, et al. [35]	The Journal of Craniofacial Surgery	2012	31	F	scalp (occipital)	Not informed	no	CNS local invasion	resection	not informed
13	Celia Requena, et al. [36]	Am J Dermatopathol	2013	82	M	glabellar	Not informed	no	iFrontal and ethmoid bone invasion	Extensive resection with local flap (skin + duramater) + radiotherapy	not informed
14	Hayato Takahashi, et al. [37]	Am J Dermatopathol	2004	22	F	halux E	2,2	sim	invasaoossea	amputação com margem de 3cm	SED 20 meses
15	James C Steinmetz, et al. [38]	Journal of the American Academy of Dermatology	1990	59	M	escápula D	4	não	linfonodal mediastinal, disseminada	biópsia excisional + ampliação de margens	óbito 9 semanas após a cirurgia
16	Hirsch and Helwig [39]			50	F	face	nãoinformado	não	não		SED 18 meses
17	Sharvill [40]	Am J Dermatopathol	1986	39	F	punho D	2	sim após 36 meses	não		sem informação após a recorrência local
18	Rosborough			83	F	braço E	3	nao	linfonodalaxilar		SED 17 meses
19	Matz, et al.			80	F	courocabeludo	5	sim	linfonodal cervical, disseminada		óbito após 84 meses
20	Schremmer			55	M	dedo	3	nao	linfonodalaxilar, pulmonar		nãoinformado
21	Hilton, et al. [41]			14	F	braço E	3,7	sim	linfonodalaxilar		SED 17 anos
22	Lucas and Nordby			74	F	mao E	nãoinformado	sim	não		SED 24 meses
23	Webb and Stott [42]			52	F	coxa D	10	sim	linfonodal inguinal		SED 18 meses
24	Botha and Kahn [43]			15	F	orelha E	nãoinformado	sim	não		SED 6 anos
25	Dissanayake and Salm [44]			79	F	sacro	8	nao	pulmonar		óbito após 1 ano
26	Harrist et al. (2 cases) [45]			33	M	pe E	nãoinformado	nao	pulmonar		perdeuseguimento

27				70	M	pe E	8	sim	linfonodal inguinal, ossea, pulmonar		óbito apos 7 anos
28	DeMoraes et al. [46]			23	F	perna D	4	nao	supraclavicular, pulmonar		nãoinformado
29	Shvili and Rothem [40]			44	F	nádegas	5	nao	linfonodal inguinal, disseminada		óbito apos 6 meses
30	Hermann, et al. [47]	Skeletal Radiologia	1987	13	F	courocabeludo	1,4	sim - 3 recidivas	linfonodal cervical, óssea		óbito apos 34 meses
31	Clark [48]	Conn Med.	1987	74	F	coxa D	8	nãoinformado	nãoinformado		nãoinformado
32	S Vohra et al [49]	The Foot	1996	39	M	Hilux D	3	nao	nao	amputa;ao parcial do halux	SED 6 meses
33	Consuelo Sa´Nchez Herreros, et al. [50]	Dermatol Surg	2011	93	F	nariz	2,5	nao	nao	cirurgia micrografia de Mohs + reconstrua;ão com retalho fronta;	SED 48 meses
34	John L Kiely, et al. [51]	Thorax	1997	50	F	mao E	nãoinformado	sim	pulmonar 17 anos apos primario	exerece do primário, sem teto das metástases	nãoinformado
35	Sun TB, et al. [52]	Journal of the Formosan Medical Association	1996	9	M	Pe D		sim 3 vezes em 10 anos	óssea, apos 1 anodisseminada	exegese do tumor, amputa;ao transitaí	óbito 36 meses apos as primeiras metástases e amputa;ao
36	Barnett MD, et al. [53]	Am J Clin Oncol	2000	34	M	Pe D		sim			
37	Medina Henriquez JA et al. [54]	Scand J PlastReconstr Surg Hand Surg.	2001	37	M	Mao		não	não	amputa;ao + retalho antebraquial reverso	SED 5 anos
38	Menéndez RH et al. [55]	J Neurosurg Spine	2015	63	F				subdural (T9)	resseccao da meta + radioterapia	SED 2 anos
39	Solomonov A, et al. [56]	Respiration	2001	65	M	parede abdominal				resseccão + radioterapia / braquiterapia para meta	boa resposta (?)
40	Kiran Mishra, and Sarla Agarwal [33]	Acta Cytologica	1998	40	F	courocabeludo	5	nãoinformado	linfonodal cervical	resseccão + linfadectomia	nãoinformado
41	Hong JJ, et al [57]	Dermatol Surg	1995	40	M	suprapubico	7	sim		exerece	

Table 1: 41 cases of malignant chondroid syringoma.

Diagnosis is made essentially by histological study. The panoramic view shows asymmetry and little circumscription, with small clusters of epithelial neoplasia at a certain distance from the main malignant mass. The tumor has an epithelial and non-epithelial component with a remarkable amount of mucin. The epithelial component is composed of tubular or solid aggregates of polygonal or plasmacytoid cells, ranging from relatively monomorphic to extremely pleomorphic. Variations in the size and shape of neoplastic cell aggregates are common, with large nuclei and abundant mitotic figures, and often some areas of necrosis. The stroma is usually myxoid, but it might present as chondroid and even osteoid [8]. Immunohistochemistry haven't played a import role on differential diagnosis due to a small number of cases studied, with variable immunohistochemical results [8].

The currently recommended treatment is surgical excision with inclusion of tumor free tissue to ensure complete tumor removal [2]. Although several reported cases have presented adjuvant treatment with chemotherapy and radiotherapy, there is no evidence of its benefit [13].

Since most of the cases described are small, circumscribed nodular tumors, the necessity for flap reconstruction is not frequent. We found in the literature cases that required local flaps due to location or dimensions and amputations of distal extremities due to local bone invasion or impossibility of free margins, without any description distal flaps requirement. However, its insidious evolution makes it prone to late diagnosis especially in countries with more difficult access to health services, which leads to complex defects requiring adequate reconstruction.

Considering upper limb reconstruction, the approach requires a stable and durable solution, and the flaps used, especially in oncological surgeries, should be well vascularized, must provide bone

coverage, control of infectious processes, resistance to possible complementary treatments and maintenance of function and esthetics of the limb. Free muscular transfers have been recommended for these reconstructions, however, their greatest technical difficulty and complications cannot be forgotten. Among the alternatives, we highlight the latissimus dorsi flap, being a technically simpler option, accessible and with low morbidity of the donor area.

The success rate with the latissimus dorsi flap for diverse reconstructions ranges from 72% to 95%. As a result of its vascularization, various shapes and sizes of this flap might be executed, depending on the location, cause and defect to be repaired. The skin island might reach dimensions up to 35 cm x 12 cm while still maintaining direct closure of the donor area. This flap can still be used in a functional way, restoring the elbow flexion or extension movement [10,12].

Conclusion

The chondroid syringoma of the upper limb usually have a late diagnosis, making extensive lesions a common presentation, which leads to aggressive oncologic surgical treatment, which results in expressive defects. Thus, the use of the latissimus dorsi flap is often used mainly because of the intrinsic characteristics of possibility of good functional and aesthetic restoration.

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