Giant cystic axillary hemangiolymphangioma. A case report

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Case Report

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Introduction: Hemangiolymphangioma is an extremely rare vascular malformation comprised of both endothelial and lymphatic components. It is a subgroup under the umbrella of endothelial malformations(1). It is believed to be caused by anomalous embryological development, leading to the formation of a rapidly growing tumor (2). The most common site of presentation occurs in the anterior and posterior cervical triangle of the neck. We present the case of a 20 year old male with an rapidly growing axillary tumor of 6 months of evolution, referring no symptoms at all. The case is explained in detail to the patient and later it is decided to intervene surgically on the person to remove the tumor.

Keywords: Hemangiolymphangioma

Introduction

Cystic hemangiolymphangioma is a congenital benign malformation of unfrequent lymphatic and blood vessels that usually occurs during childhood and is very rare in adults(1). 40–60% of HLAs are discovered at birth, 80–90% during the first 2 years of life, and decreases in frequency with age (1). It can be located anywhere in the body, but especially in the area of the head, neck, armpit, oral and maxilofacial region, abdominal cavity such as duodenum, colon, bladder, testis and vertebral column. Tomography and ultrasound remain useful diagnostic methods and complete surgical resection(2) continues to be the treatment of choice(3)(4). The clinical presentation is further complicated as HLAs are associated with a multitude of syndromes (1). As a result, the differential diagnosis includes congenital (branchial cleft cyst, thyroglossal duct cyst—most common, vascular anomalies, laryngocele, ranula, thymic cyst, etc.), inflammatory (infectious and noninfectious disorders), and neoplastic (squamous cell carcinoma—most common, lipoma, thyroid masses, etc.)(1).

Case report

This is a 20-year-old male, with no surgical history, occasional alcoholism and positive drug addiction to marijuana use 1/2 cigarette daily from the age of 16. He goes to the outpatient service for presenting left axillary tumor of 6 months of evolution. Upon interrogation, he mentions progressive increase in volume in the absence of trauma, until reaching the clavicular midline of euthermal, painless, asymptomatic characteristic, without change of coloration. External ultrasound (US) report of soft tissues shows complex cystic tumor; beginning with a tumor approach under study located in the left axillary region. Computed tomography (CT) of the single neck is requested, and lesion of unspecified etiology is reported. Subsequently, simple thoraco-abdomino-pelvic CT scan is performed and contrasted intravenous and oral to evaluate the extent of the lesion, evidencing cystic lesion in the left axillar region in its maximum axis of 6.3 x 16 x 16 cm, to consider giant cystic lymphangioma as a diagnostic probability so it is scheduled for diagnostic surgery.

Lymphangioma resection was performed, dissecting until vein and axillary artery were found. The piece is extracted, irrigating with sterile solution, performing hemostasis and closure by planes, without evidencing transoperative findings. After completing the first postoperative day without eventualities, it is decided to discharge from the service with follow-up by external consultation.

The hepatological examination reports a piece of 370 g, with measures of 19 x 11.3 x 4 cm with histopathological diagnosis of hemangiolymphangioma.

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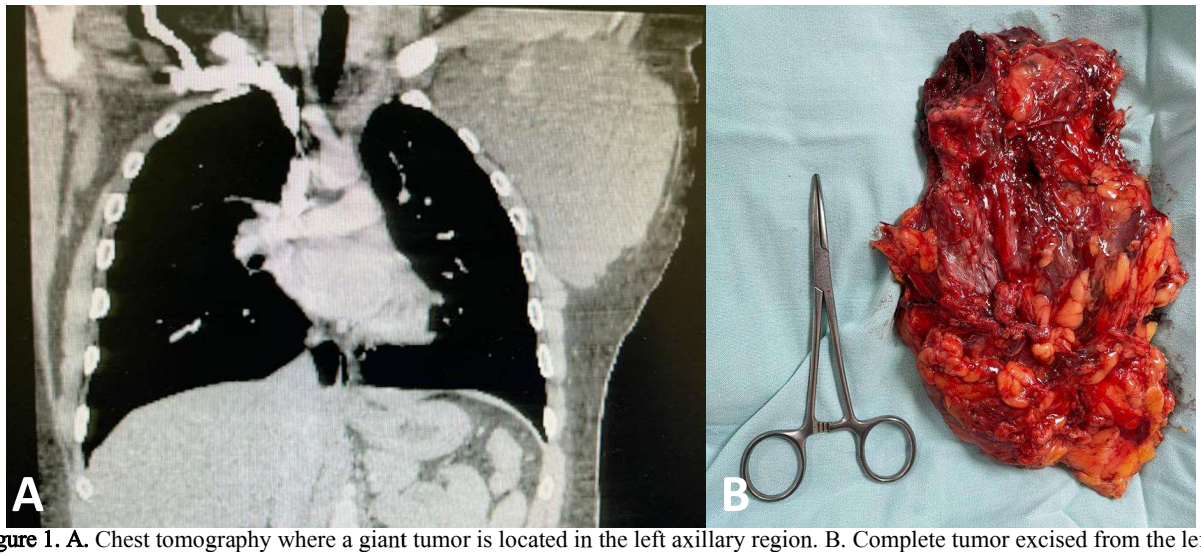

Figure 1. A. Chest tomography where a giant tumor is located in the left axillary region. B. Complete tumor excised from the left axillary region

Discussion

Hemangiolymphangioma is considered to originate from a process of altered angiogenesis caused by an excess of fibroblast growth factor (2). It affects more frequently the head and neck region and between 80% of the cases it affects female (2). Vascular malformations are classified by the vessels present, such as arterial, arteriovenous, venous, capillary or lymphatic malformations. Mixed vessels encompass congenital lymphangiomas or lymphatic anomalies which are aggregates of lymph vessels (6). When lymphangiomas are filled with blood, they are referred as a mixed hemangiolymphangioma, which is an extremely rare entity (1). Unlike hemangiomas which proliferate, vascular malformations enlarge in proportion to the growth of the individual and expand by hypertrophy. In most cases it´s not visible at birth or early years of life, but they may present rapid growth at later stages of life, becoming more evident after infection, injury or trauma. The so called "early ones become evident during development and the late ones appear during adult life (3). The doppler ultrasound is an excellent method of diagnose when the tumor is just beneath the skin, but when the lesion its deeper or affects abdominal cavity it´s better to diagnose the tumor with a CT scan or MRI. Surgical management is the first alternative, although it has a recurrence rate of 17% when the macroscopic resection is complete and 40% if it´s incomplete. Complete removal can only be performed in three quarters of them, the rest compromise vital structures that require a partial resection (3).

Conclusion

Hemangiolymphangioma are malformations formed by lymphatic and blood vessels. It is believed that they may be caused by the anomalous embryological development of the lymphatic and vascular system, which leads to the formation of a fast-growing tumor (2), however, there is evidence of cases of hemangiolymphangiomas with presentation in adulthood, apparently secondary to trauma in areas with abnormal lymphatic vessels(2). The diagnosis can be clinical; however, for loculated or deep lesions it is identified and there is better surgical planning by performing a computed tomography or better with an MRI. (4) There are several therapeutic options; the definitive treatments are surgical resection. (3)(4). Sclerotherapy may be tried as an alternative treatment in case of recurrence (6).

Conflicts of interests

There are no conflicts of interest for this publication.

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