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Hemangiolymphangioma in an adolescent: Imaging implications

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Background

A lymphatic-venous malformation (LVM) also referred as "Hemangiolymphangioma" is a lesion that contains mixed venous and lymphatic components. This entity may affect multiple parts of the body, most commonly the head and neck; however, LVM is rarely reported in the English literature within the mediastinum [1,2]. Mediastinal masses may be secondary to several etiologies, and their characterization can be complicated due to the existence of an ample number of differential diagnoses to consider [3]. The etiology of LVM is thought to be embryologic in origin since the lymphatic system developed in relation to the venous system [4]. Clinical presentation of LVM involving the mediastinum range from asymptomatic patients, to chest pain and dyspnea mainly due to mass effect on nearby vasculature and nerves [1]. LVM is important to recognize in a rapid manner because the management and treatment depends on the prompt diagnosis obtained by the imaging characteristics and the pathology report [1]. In this case, we present a fifteen-year-old patient with a LVM affecting the mediastinum. The LVM mass was subsequently treated with surgical resection.

Description

A fifteen-year-old Hispanic male presented to the emergency department with precordial chest pain, fever, and weakness. Chest radiograph demonstrated cardiomegaly (Figure 1). An echocardiogram was performed and revealed pericardial effusion and suggested pericarditis. Computed tomography of the chest with intravenous contrastmaterial revealed serpentine contrast filled structures within the low attenuated mediastinal mass (Figure 2). Subsequently, a pericardial window was performed, and tissue biopsy was obtained. Surgical resection was the decided treatment, which led to the removal of a brownish mass from the mediastinum (Figure 3). Pathology report showed irregular vascular and lymphatic spaces that are anastomosed and present different calibers. Benign endothelial cells infiltrating the surrounding adipose and fibrous tissue. Findings were consistent with hemangiolymphangioma and pericarditis (Figure 4-6).

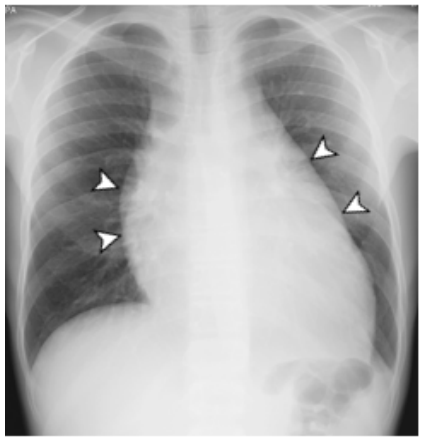

Figure 1: Chest radiographs, Poster-anterior view demonstrated cardiomegaly (arrowheads).

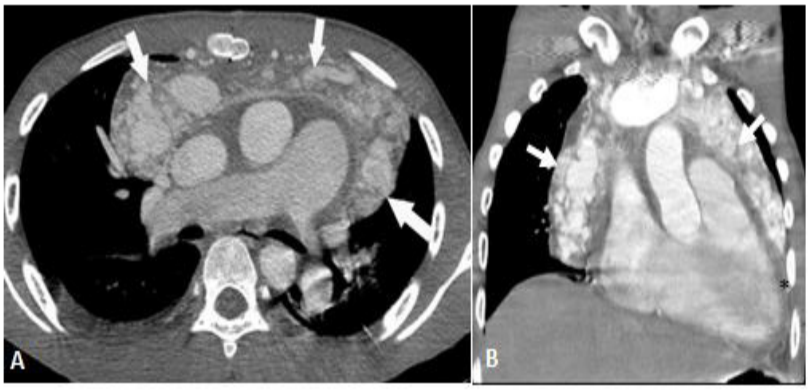

Figure 2: Computed tomography of the chest with contrast. A. axial, B coronal planes revealed tortuous contrast filled structures within the low attenuated mediastinal mass (white arrows). There is a scarce pericardial effusion (black asterisk) and dilation of the right sided heart structures. Findings were consistent with Hemangiolymphangioma

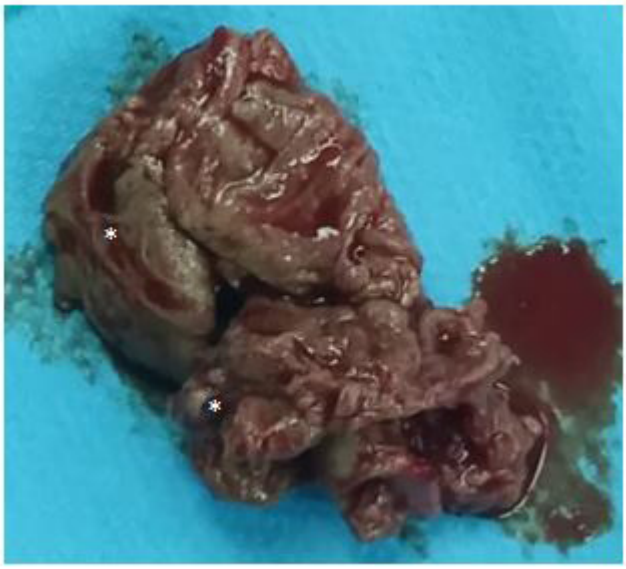

Figure 3: Surgical resected brownish mass with vascular components (white asterisks) compatible with Hemangiolymphangioma.

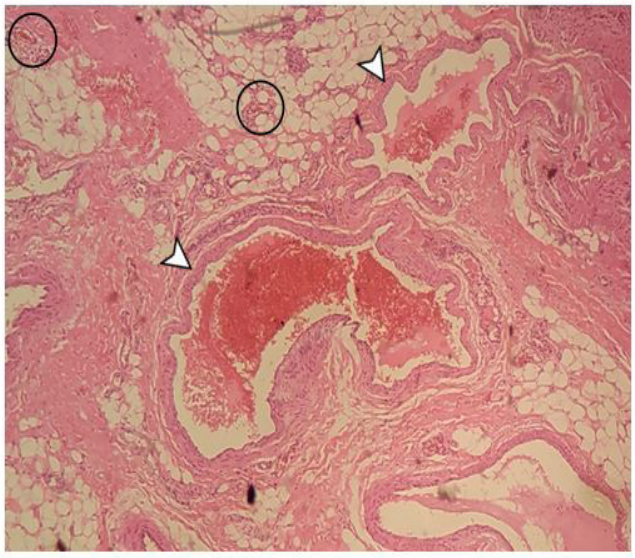

Figure 4: H&E 40x. Benign mesenchymal neoplasia with vascular differentiation formed by dilated vessels (arrowheads), which are irregular and surrounded by smaller vessels (circles).

Discussion

Vascular lesions are varied in nature and etiology. The international Society for the Study of Vascular Anomalies (ISSVA) classified these lesions into vascular malformations and vascular tumors. Vascular malformations include combinations of venous, arterial, lymphatic, and capillary components [5]. Lymphatic-venous malformation are low flow lesions in comparison to arterial malformation which demonstrate high flow [6]. Since LVM were first reported in 1983, imaging has been a hallmark of the diagnosis. Symptoms ranged from asymptomatic patients to severe dyspnea secondary to airway obstruction or stroke like

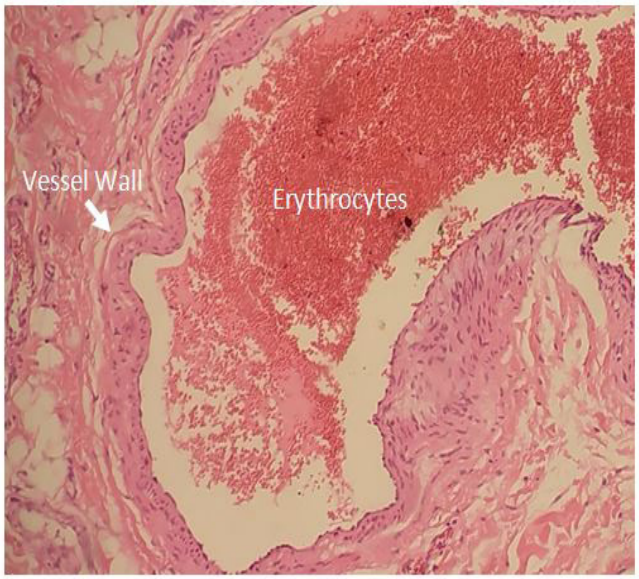

Figure 5: H&E 100x. Venous component with complete layers and tunica included.

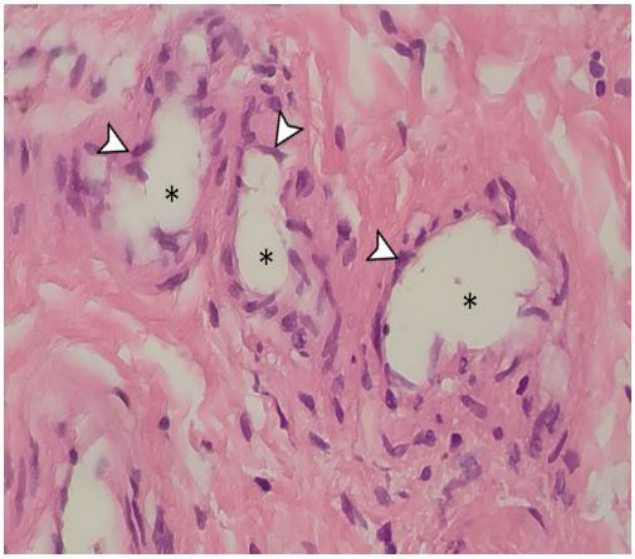

Figure 6: H&E 400x. Lymphatic component in clusters. Made from prominent endothelium (arrowhead).Lumens do not contain erythrocytes (black asterisks).

symptoms [1,6]. Throughout the years, computed tomography has been used frequently in the diagnosis of LVM to elucidate whether they are close to structures that make them unresectable, or to understand where they are obtaining their blood supply. Most commonly, LVM draining to the superior vena cava or the brachiocephalic vein [2,7,8]. M Xu et al stated that the aforementioned vessels are not only a drainage for LVM, but also the supply [9]. On computed tomography (CT) scan, LVM demonstrate homogenous low attenuation with serpentine contrast filled structures representing the malformation between lymphatics and veins. Since LVM are low-flow lesions, thrombosis with subsequent calcification may arise as a complication [10]. Multiplanar CT reconstructions is used to better characterize venous connections to LVMs [9]. Magnetic resonance imaging use is crucial to differentiate high versus low flow lesions since slow flowing venous blood may showed high or intermediate signal intensities on T2 or T1-weighted images respectively. Lymphatic components of the LVM reveal high intensity signal on T2-weighted images [2,11]. Some areas of high signal intensity are observed secondary to thrombi and fat composition within the LVM. Moreover, scattered areas of low signal intensity are likely due to phleboliths [10]. Even though these masses can be imaged by several modalities, the final diagnosis is performed by pathology [2]. It is important to note that LVM may have varied ratios of venous and lymphatic components, which may result into atypical imaging findings such as absence of calcifications and subtle enhancement [4,12]. Apart from the initial diagnosis, it is essential to know the anatomical structures surrounding the LVM. This is critical to decide the type of treatment to provide. The mainstays of treatment for LVM depend on symptomatology and how resectable the lesions are [13]. Management for LVM ranged from observation to sclerotherapy and surgical resection [1]. Complication of the surgery include hemorrhage from a vessel within the mass or damage to surrounding mediastinal structures such as the phrenic nerve [2,14,15].

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