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A Case of Arteriovenous Haemangioma in the Neck Masquerading as a Brachial Cleft Cyst in a Young Adult

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Abstract

Arteriovenous haemangiomas are a rare entity and it constitutes 7-10 % of all soft tissue lesions. They usually present as swellings with skin discoloration and may be associated with mass effect. Given history coupled with imaging are usually sufficient concluded the diagnosis. This is an 18 years old teenager who presented with a history of neck swelling the last 2 years progressively increasing in size with no functional deficit or mass effect. Though arteriovenous haemangiomas are rare, it must be included in the differential diagnosis. Hence a thorough knowledge on the nature of the condition is required, in order to diagnose and manage it efficiently.

Keywords: Arteriovenous haemangioma; Acral haemangioma; Branchial cyst

Abbreviations: MRI: Magnetic Resonance Imaging; CT: Computed Tomography; ISSVA: International Society for the Study of Vascular Anomalies; ICA: Internal Carotid Artery; ECA: External Carotid Artery.

Introduction

Neck masses differential diagnosis in adolescents is broad and exhaustive. It can be classified in many ways following the anatomical location, neoplastic, embryology and etc. A thorough history is important in making a preliminary diagnosis and its subsequent management. However, despite the initial history, imaging and histopathology, the final diagnosis could be a diagnosis which is not expected.

Hemangiomas represent approximately 7-10% of all benign soft tissue masses [1]. Arteriovenous haemangioma is also known as arteriovenous tumour. The pathogenesis of this lesion is unknown. However, hamartomatous proliferation either of the subpapillary vascular plexus or of the Sucquest -Hoyer canal of the true glomus is proposed as a possible histogenic mechanism [2]. Given the location of the lateral neck swelling and the age of presentation of this patient, branchial cyst was the initial diagnosis. He is an 18 year old adolescent who presented with a slow growing lateral neck swelling for the past two years. He initially sought treatment at a district hospital before being referred to our tertiary centre for further management.

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Case Presentation

An 18 year old adolescent, who was well prior to this, was referred to the plastic and reconstructive team for a left supraclavicular swelling which was increasing in size for the last two years (Figure 1). It is occasionally associated with pain. He had no prior history of trauma.

Figure 1: Preoperative marking over the lateral neck swelling.

Upon examination, the skin over the mass appears normal with no dermatologic manifestations. No thrill palpable on palpation. It is a soft and mobile swelling and the borders are not well demarcated. It measures about 10 cm x 10 cm. No initially FNAC or biopsies were taken. A computed tomography (CT) scan which was done two years ago revealed a well-defined hypodense lesion (HU9-30). It is multilobulated and measures 5.3 x 7.4 x 8.3 cm (AP X W X CC) with thin wall peripheral enhancement. It occupies the subcutaneous fat and is bounded anterosuperiorly by left sternocleidomastoid muscle, medially by left scalenus anterior, scalenus medial muscle, left jugular vein, and left common carotid artery, posteriorly by left trapezius and levator scapulae muscles, laterally by left trapezius muscle and inferiorly by left subclavian artery and vein. This cystic mass insinuates in between the neck structures without causing much mass effect. A discussion with the radiologist after reviewing the initial CT scan led to the decision to proceed with a magnetic resonance imaging (MRI) scan in order to reevaluate the mass. The MRI revealed an enlarging mass which is hyperintense in T2WI and STIR and hypo intense in T1WI and was suggestive of a second branchial cyst. He underwent excision of the left neck swelling and intraoperatively the mass was excised in total (Figures 2&3). Mass clinically looks lobulated and soft measuring about (10cmx8 cm). Primary closure of the skin was obtained without tension. The mass was sent to the pathologist for examination.

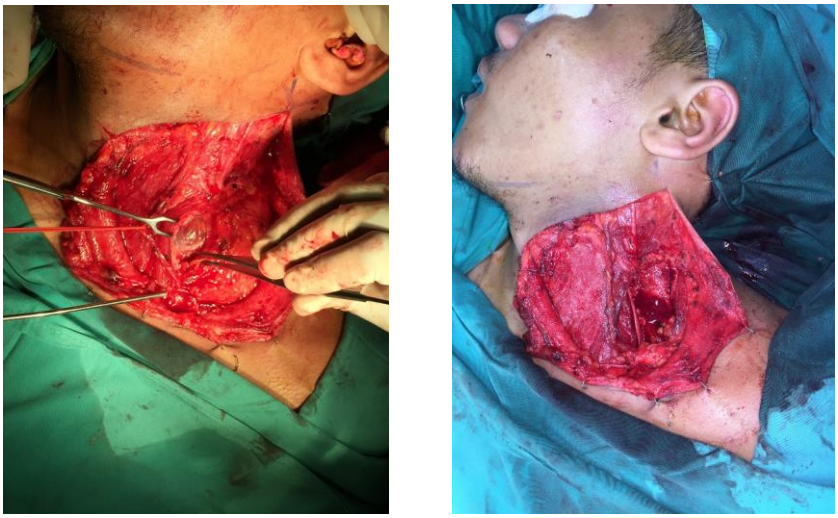

Figure 2 & 3: Intraoperative pictures of arteriovenous hemangioma and post excision.

Cut sections of the cystic lesion shows a multilobulated cyst which contains straw colored fluid. Histologically, sections from the tissue found fibro collagenous tissues composed of multiples vascular spaces with thick and thinned walls lined by flattened endothelial cells. There was no dysplasia or evidence of malignancy seen and it was reported as an arteriovenous hemangioma (Figure 4).

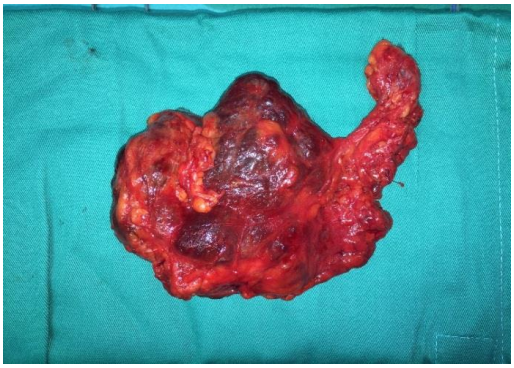

Figure 4: Excised specimen.

Discussion

Arteriovenous hemangiomas or also known as arteriovenous tumours or acral arteriovenous tumours, are a distinct, benign, acquired vascular lesion, and was first reported by Baberstein and Jessner in 1956 [3]. There are two distinctive variants which are the deep seated and cutaneous (acral arteriovenous tumour) type [2]. The so-called "deep" type occurs in younger individuals, is associated with varying degrees of shunting, and is regarded as a malformation. Although called "deep", lesions can occur close to the skin and they may pulsate or writhe due to afferent arterial blood flow if large shunts are present. These lesions are referred as cirsoid or arteriovenous aneurysms. The superficial form includes lesions classified as arteriovenous hemangioma/shunt by Ciirard et al and as acral arteriovenous tumour by Capareto et al [2]. The International Society for the Study of Vascular Anomalies (ISSVA) classification of vascular anomalies has classified acral arteriovenous tumour as provisionally unclassified vascular anomalies [4]. Ultrasonography, CT scan and MRI and Doppler can be a valuable tool in providing a preliminary diagnosis. This must be coupled with a comprehensive history as many imaging findings can overlap with thousand over diagnosis. Preoperative imaging is vital in guiding the operative strategy aimed at the complete removal of the lesion and also to help reduce surgical complications.

In this patient, he was not sure if the lesion was present when he was younger but for the past two years he noticed that it was slowly increasing in size since he was 16 years old. With the given history, a branchial cyst would be one of the most probable diagnoses especially since the lesion occurred on the lateral neck, anterior to the sternocleidomastoid muscle. On CT scan, branchial cysts can be described as rounded or spherical, sharply circumscribed, with fluid density centrally surrounded by a thin wall. Extension of the cyst wall between the internal carotid artery (ICA) and external carotid artery (ECA) just above the carotid bifurcation has long been established as a pathognomonic sign. On MRI, branchial cysts show variable signals in T1-weighted images, high signals in T2-weighted images and no enhancement in uncomplicated cases in T1 C+ (Gd) images. However, hemangiomas on the other hand, may appear as an ill-defined mass of similar attenuation to muscle in unenhanced CT. CT may also show the presence of associated phleboliths. In MRI, hemangiomas are typically well- defined, lobulated and heterogeneous with no features of local invasion. While many sequences show a rather heterogeneous signal mass, certain signal characteristics tend to dominate. T1-weighted images signal is often intermediate to slightly high (relative to skeletal muscle), and some focal high signal areas may be present in a large proportion of lesions (up to 70%). High signal intensity tends to dominate on T2- weighted images. On T1 C+ (Gd) images, lesions show marked signal enhancement in parts of the areas, which were both of high and low on T2-weighted images [5].

Arteriovenous hemangiomas can rarely present with normal skin without any discoloration, venous engorgement or inflammatory signs. Keeping this in mind, any swelling that is associated with pain should be investigated radiologically before proceeding with a biopsy. This will give a preliminary idea to the pathologist as to what he/she is dealing with – either or a vascular lesion as in our case – and thus help avoid a delay in diagnosis and any possible complications associated with it [6]. Histologically, arteriovenous haemangiomas are characterised by multiple thick and thin walled vascular spaces resembling arteries and veins, respectively. [2] On the other hand, more than 90% of branchial cleft cysts are lined by stratified squamous epithelium which may or may not be keratinised. The wall of the cyst typically contains lymphoid tissue - germinal centres formation. Some authors have noted the presence of hair follicles and sebaceous and sweat glands within the cyst [7]. The mainstay treatment of arteriovenous haemangiomas and branchial cysts is via surgical excision. As the neck harbours many vital structures, surgical excision of lesions in that area must be done with utmost care as many vital structures may lie adjacent to it. Both lesions can recur if inadequately excised.

Conclusion

Arteriovenous haemangiomas although uncommon should be included in the differential diagnosis of any neck swelling. The uniqueness about this case is that haemangiomas does present with normal skin without cutaneous changes, venous engorgement or inflammatory

signs though it's rare. It is important for the surgeons, pathologists and radiologists to be using similar classification to avoid any miscommunication, delay or inappropriate treatment.

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