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Anterior Jugular Vein Aneurysm Associated with Neurofibromatosis Type 1

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Abstract

Anterior jugular vein aneurysm is a very rare condition. Such lesions may present to the primary care and the workup should include an ultrasound with duplex. Surgical excision is the accepted treatment for patients with cosmetic deformation or complications. In our case the patient with Neurofibromatosis type 1 and suspicious thyroid nodule suffers from intermittent neck swelling is presented.

Keywords: Aneurysm; Anterior jugular vein; Neurofibromatosis

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Introduction

Venous aneurysm is an abnormal dilation of vein which needs to be considered in the differential diagnosis of neck swellings.

Venous aneurysms have been reported in several anatomic sites in the neck, the typical location being the internal and external jugular veins and the commonest is the internal jugular vein [1-8]. Anterior jugular vein aneurysms are extremely rare with a few cases been reported in the English literature [2-14]. Aneurysms can be classified by type (true and false), morphology (saccular and fusiform), etiology (congenital and acquired), and location.

We report a case of a 59 year old woman with left anterior jugular vein aneurysm suffering also from Neurofibromatosis type 1 and a suspicious thyroid nodule.

Case Report

A 59 year old woman presented with prolonged history of a soft, non-pulsatile, non-tender, intermittent swelling in the anterior left neck that appeared and became more prominent with Valsalva maneuver or during talking, and resolved after rest. There was no history of trauma or surgery. Medical history-neurofibromatosis type I and right leg angioma excision in the past (**Figures 1 and 2**).

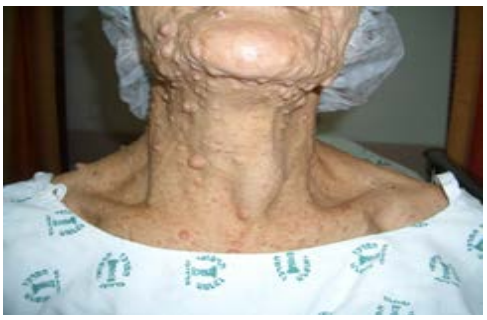

Figure 1 Photograph of neck with the patient performing a Valsalva maneuvere. A swelling of the anterior neck is easily seen. In addition, multiple neurofibromas are observed in the neck and face.

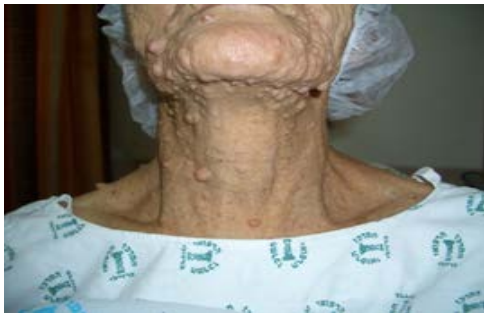

Figure 2 Photograph of neck with the patient at rest. There is no swelling as seen on Valsalva, multiple neurofibromas are observed in the neck and face.

Ultrasound examination (US) of the neck revealed a 3.3 cm hyperechoic cystic lobular nodule with poor peripheral vascularization in the thyroid isthmus and 0.8 cm hypoechoic nodule with poor peripheral vascularization in the right lobe of the thyroid gland. There was no vascular abnormality on the US.

Venous duplex scan showed anterior jugular vein dilation during

Valsalva maneuver up to 2.83 cm. The differential diagnosis according to the US was a vascular malformation vs an aneurism.

Contrast enhanced computer tomography scan revealed vascular and hypodense thyroid lesions correlating with US (**Figures 3 and 4**).

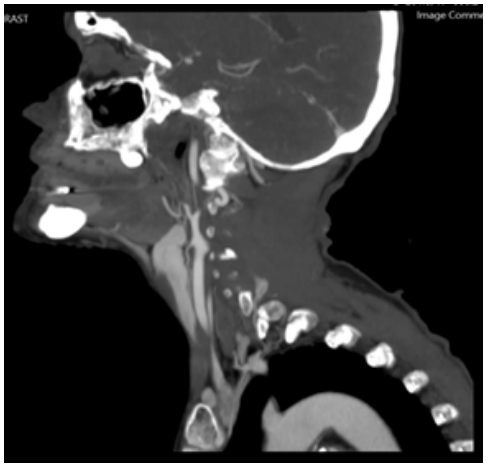

Figure 3 Computed tomography with contrast of the neck sagittal view depicting an aneurism of the anterior jugular vein. The internal jugular vein is observed as well with a small connection between the veins.

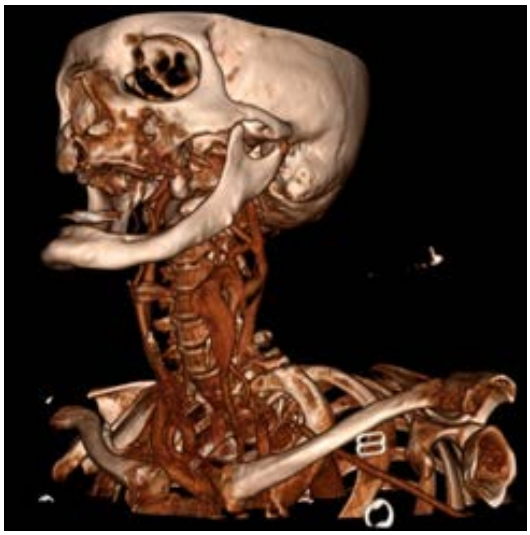

Figure 4 Computed tomography reconstruction of the venous system. The aneurism is easily depicted along the upper part of the anterior jugular vein

Fine needle aspiration (FNA) from the thyroid nodule showed Hurtle cells with varying degrees of atypia. Right hemithyroidectomy with isthmectomy and anterior jugular vein aneurysm excision were performed. At surgery the diagnosis of the aneurysm was confirmed. Histopathology of the anterior neck mass revealed aneurysmal dilatation of vein wall and of the thyroid nodule showed extensive Hurtle cells differentiation and focal atypia (**Figures 5 and 6**).

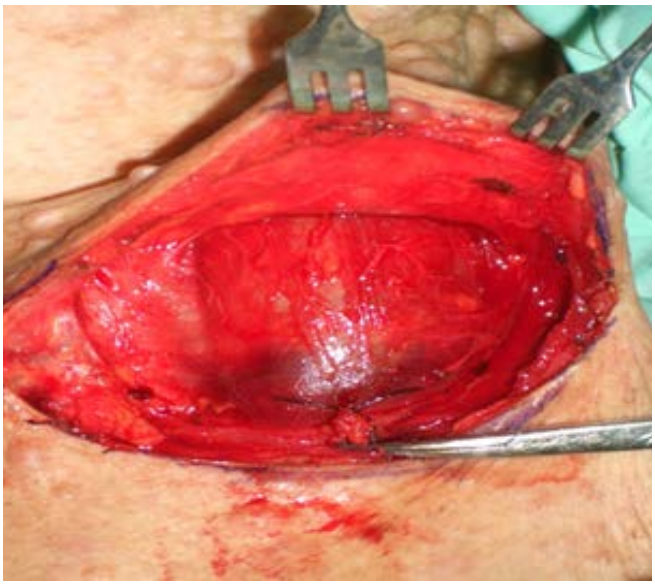

Figure 5 Photograph of neck swelling after ligation of the left anterior jugular vein below the aneurism.

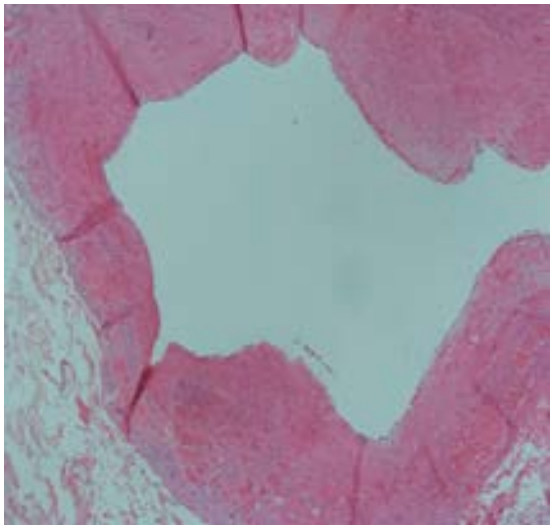

Figure 6 Aneurysmal dilatation of vein wall on the histopathology

Discussion

Venous aneurysm refers to localized abnormal dilatation of vein. According the etiology, venous aneurysms can be primary (congenital) or acquired due to trauma, surgery, infection, or can appear spontaneously with no etiologic cause identified [15]. Complications associated with venous aneurysms include: thromboembolism, rupture, venous obstruction, compression of adjacent tissues. According to the morphology, aneurysms are divided into saccular and fusiform. The internal and external jugular veins are generally affected. The internal jugular vein is the commonest location of the disease; the external jugular vein is the second commonest site of involvement [1-8]. Only a few cases of anterior jugular vein aneurysm have been reported in the English literature [2-14]. In our case it was a saccular non-

complicated aneurysm which appeared spontaneously with extremely rare location in the anterior jugular vein.

Venous aneurysms of the head and neck region most often present as asymptomatic soft tissue masses that increase in size with a Valsalva maneuver. A diagnostic venous hum is rarely heard [16,17].

Differential diagnosis soft tissue cervical masses with enlargement during Valsalva maneuver include laryngocele, venous aneurysm and superior mediastinal cysts [2,12,18].

Diagnostic studies employed in the evaluation process include duplex ultrasonography, venography, CT and MRI scans. In our case US with duplex, CT scan were performed and the duplex scan diagnosed anterior jugular vein dilation during Valsalva maneuver up to 2.83 cm.

The association of venous aneurisms and neurofibromatosis is considered rare [19-23]. The relation between vascular anomalies and neurofibromatosis has been studied. Neurofibromin expression has been demonstrated in the vascular endothelial and smooth muscle cells. Deficiency in in this protein may lead to proliferation of the cells in the vessel wall similar to the proliferation of peripheral nerve cells [24,25].

However venous aneurisms in association with neurofibromatosis type 1 are very rare and the few reported cases were from the internal jugular vein. Hence this is the first report to our knowledge of an anterior jugular vein aneurism associated with neurofibromatosis.

The indication for surgical treatment of a venous aneurism is cosmetic complaints or complications such as hemorrhage or venous thrombosis. In our case the indication for treatment was cosmetic. Furthermore, since the patient was planned for a partial thyroidectomy the risk of treating the aneurism was minimal.

Conclusion

Anterior jugular vein aneurysm is a very rare condition. Such lesions may present to the primary care and the workup should include an ultrasound with duplex. The diagnosis of a vascular lesion is considered when the mass enlarges during the Valsalve maneuvere. Surgical excision is the accepted treatment for patients with cosmetic deformation or complications. In our case the patient suffers also from Neurofibromatosis type 1. delivered.

Acknowledgement

None

Conflict of Interest

None

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