Case report

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Pseudoaneurysma of the external jugular vein communicating with the internal jugular vein. Case report.

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Abstract

True and false aneurysmas (pseudoaneurysmas) of veins are very rare conditions. We present the case of a 58 year old woman with a progressive spheric enlargement with pusatile character of the right laterocervical area. Ultrasonography established the diagnosis showing the vascular mass with turbulent flow belonging to the external jugular vein and the communication with the internal jugular vein. The particularities of this case are the absence of an obvious etiology and the particular variant of flow direction from internal towards the external jugular vein. To the best of our knowledge, this is the first case of pseudoaneurysma of the external jugular vein reported in literature.

Keywords: false aneurysm, jugular vein, Doppler ultrasonography

Introduction

The first venous aneurysma was described by Harris in 1928 [1]. Aneurysmas are frequently connected to arterial vessels; due to the low pressure in the caval system, they are seldom encountered in veins. There is no specificity for age, gender, or localization, the most common affected veins being the external (EJV) and internal (IJV) jugular veins [2]. Only a few authors have described till now aneurysmal dilatation of the EJV [2-12].

Aneurysma of the EJV may be classified in primary (congenital) and secondary (post-traumatic) aneurysmas; the former can affect more frequently the neck and chest [13]. The most common lesions after failing to locate the IJV in central venous catheterization are pseudoaneurysmas (PA), or false aneurysmas [14].

In this case report we described a patient with PA of the right EJV.

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Case report

A 58 year old woman, with a personal history of primary arterial hypertension, chronic atrial fibrillation, mitral and aortic regurgitation, and moderate pulmonary hypertension, was admitted for her routine cardiological follow-up, complaining of cervical constriction with pusatile character. In the last year, she had noted the progressive spheric enlargement of the right laterocervical area, this phenomena being connected to physical and defecation effort. Additional, she complained of a local pulsatile burning sensation, independently of physical activity or position. She had no history of local trauma or autoimmune systemic disease.

Clinical examination of the cervical region revealed superficial, spheric enlargement of the right cervical area. This enlargement became larger during Valsalva maneuvre, diminished in orthostatism (fig 1), or disappeared completely after a little digital pressure of the neck Pal-



Fig 1. a) Enlargement of the laterocervical mass during bending (arrow). b) Disappearance of laterocervical mass in orthostatism

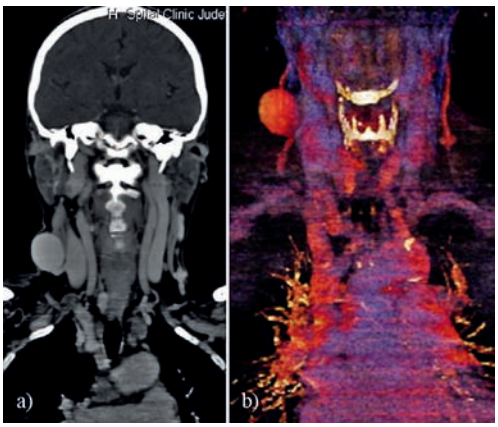


Fig 3. a) Grey-scale Angio-CT (frontal section): Pseudoaneurysma with the collum connecting to the IJV. b) Colour Angio-CT: Pseudoaneurysma of right external jugular vein.

pation revealed a 3 cm diameter soft reducible, mobile, and insensitive mass, and no locoregional adenopathies. The cardiovascular examination showed complete arrythmia (atrial fibrillation) 80/min and blood pressure of 120/80mm Hg. Apart from these, the clinical examination was in normal range.

No inflammatory syndrome was present at laboratory tests and INR was 2.68 (the patient being under oral anticoagulant therapy).

Echocardiography revealed dilated left atrium and right chambers, mitral regurgitation grade II/III, tricuspidian regurgitation grade III, ejection fraction of 40%, diffuse hypokinesia of left ventricle, and pulmonary hypertension (57mm Hg).

Grey-scale ultrasound (US) described in the right laterocervical area a superficial hypoechoic structure, consisting of a vascular compressible dilatation of 3.5/3cm, with a spontaneous echoic "cigarette-smoke" sign inside, belonging to the EJV. Spectral US revealed the venous pattern. Colour Doppler US showed a turbulent flow inside the vascular mass and a communication to the IJV with flow direction from the latter to the EJV (fig 2). The US diagnostic suspicion was of PA of EJV.

Cervical angio-computed tomography (CT) revealed an enlarged right IJV (19 mm) and a well defined dilatation (3.5/3.2/3cm) belonging to the EJV, located 2 cm from the junction of internal-external jugular veins, with a collum of 5 mm. The PA was described as being located subcutaneous superficial, posterior from the EJV and lateral from the IJV; late scans showed homogeneous filling (fig 3), and no signs of complication (thrombosis or rupture).

Surgical removal of the PA was recommended, but the patient postponed the intervention. In a follow-up pe-

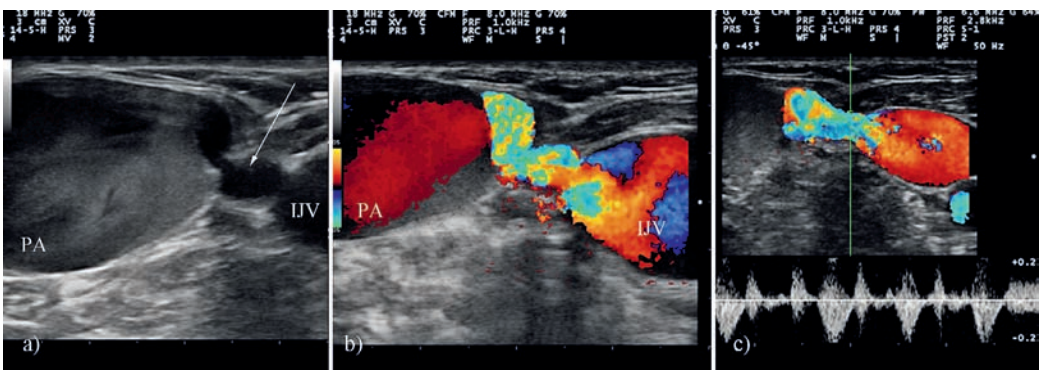


Fig 2. a) Grey-scale US: Pseudoaneurysma (PA) with spontaneous "cigarette-smoke"sign and communication (arrow) with the internal jugular vein (IJV). b) Colour-Doppler US: communication between PA-IJV with flow turbulence. c) Spectral US: communication between PA-IJV with venous pattern inside

riod of 2 months, the patients remained stable, with conservative treatment, consisting of avoidance of excessive physical efforts.

Discussion

PA of the EJV is a rare condition. Although sometimes phlebectasis (Ph) of jugular veins seems similar/ synonymous to aneurysma [15], there are several differences between them, regarding the: etiology, histopathological aspect, flow characterization, and treatment options. Congenital appearance, fusiform aspect [16], with thinned muscular layer and patent flow [12,17,18] suggest Ph. An acquired post-traumatic condition or venous diseases, saccular aspect [19], degenerative changes at histology and predilection for thrombosis suggest aneurysma [17,20]. There are also exceptions from the above rule: saccular lesions in childhood and fusiform aspects, especially in EJV [21] in adults [22]. A simple definition of these two terms regarding the veins was given by Gerwig [22], who named Ph any solitary fusiform dilatation, and Abbott and Leigh [23], who mentioned that any saccular lesion might be a true venous aneurysma. Our case seemed to have a saccular form and although there was no history of local trauma, it fitted better in the aneurysmal type rather than in Ph.

The etiology of venous jugular aneurysma include: tumors, thoracic outlet syndrome and trauma, systemic inflammatory disease, increased pressure in the venous system; the latter could be suggested in our case, as the patient had global heart failure, the other conditions had been excluded.

Histopathological analysis [5] revealed several defects in this disease: thinning/loss of elastic and muscular layers [17,24], endophlebosclerosis, endophlebohypertrophy [17] and thinning of the vein wall without any congenital anomaly. Among these, thinning of elastic layers seems to be the most important cause in congenital venous fragility [25]. As our patient postponed surgical treatment, the exact underlying histological defects were unable to be established in our case.

Diagnosis can be established by US and Doppler methods, Angio-CT scans and magnetic resonance-phlebography [26]; although multidetector-Angio-CT and selective phlebography lead to a high diagnostic accuracy, US and colour-Doppler is considered to be the gold-standard [16,27]. Hopsu E. et al described one case of aneurysma of the EJV, where US had not been useful, suggesting an infected cervical cyst or necrotic lymphnode [12]. Colour–Doppler-US established in our case the positive diagnosis of PA of EJV and Angio-CT confirmed all the aspects described by US.

Differential diagnosis include: lymphocele, cavernous haemangioma, hygroma, laryngocele, lymph node, thyroid tumor, thyreoglos/dermoid/branhial cysts, malignant transformation of a neck cyst and metastases from squamous cell carcinoma [28].

Complications of venous aneurysma include pulmonary thrombembolism, trombophlebitis, rupture, and thrombus formation. Neck and facial venous aneurysma are often asymptomatic and thrombembolic events or ruptures are less frequent [10,12,29]; although, due to the low incidence of this pathology, these risks should be taken into consideration. Ioannou et al [30] reported one complicated case of pulmonary embolism caused by an aneurysma of EJV. Our case must be closely followed-up, as the presence of the spontaneous "cigarette-smoke" sign inside the PA at US is an indicator for slow venous flow that might be a risk of thrombus formation.

Recommendations in patients with saccular aneurysma of jugular veins consist of surgical removal [31,32], while conservative treatment is considered properly in Ph [22,33-35]. In this latter variant, resection is an option, only if symptoms or enlarging/disfiguring effects appear. Other authors, such as Calligaro KD et al [10] suggested that surgery should be an option only for symptomatic and enlarging jugular aneurysma, otherwise surgery should be recommended only for aneurysma of the abdomen and lower limb.

To the best of our knowledge, this case of PA of EJV is the first to be reported in the literature. The particularities of this case were the absence of an obvious etiology and the particular variant of flow direction from IJV towards the EJV; the rare variant of the opening of the EJV into the IJV is known to occur seldom [36,37].

Conclusions

PA of EJV is a very rare condition, which can be easily diagnosed by Doppler-US with a high accuracy rate when performed by experienced physicians, other imaging methods being frequently unnecessary.

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