Internal Jugular Vein Pyogenic Capillary Hemangioma: A Case Report

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Internal jugular vein hemangioma, also called pyogenic granuloma, is a rare tumor. Such a neoformation was accidentally discovered and excised in a middle-aged man. Histologic and immunohistochemical investigations were performed, and this case is compared with the poor amount of similar ones described in the literature.

Intravascular primitive tumors are very rare diseases and few data concerning these pathologies are described in the literature. Moreover, the few cases reported are mostly focused on large veins.[1] Among these tumors, internal jugular vein (IJV) hemangioma is one of the rarest. Incidentally, it has to be mentioned that the terms capillary pyogenic hemangioma, lobular capillary hemangioma, and pyogenic granuloma (PG) are synonyms.[2]

These lesions are typically visible in the superficial derma. They are a common vascular proliferation that can rise after a minor trauma or a cutis infection and, although rarely, these lesions can form also subcutaneously and/or intravenously.[2]

The cited neoformations are very often localized in the upper part of the body, and, in particular, in arms, forearms, wrists, fingers, and, above all, neck,[3] whereas only 2 cases have been reported, respectively, in the iliac vein[4] and ovary.[5] Lesions can be associated with palpable swellings, and this could suggest a malignant event. Consequently, a correct differential diagnosis is very important to be reached.[6] Anyway, PGs show benign evolution, can be even totally asymptomatic and accidentally discovered during investigations focused on other pathologies.[7] This is what happened for the case described here.

CASE REPORT

During an echo examination focused on thyroid, a neoformation was accidentally discovered inside the left IJV of a 51-year-old man. The patient was asymptomatic, without palpable masses and he did not refer to had suffered locoregional traumas. Anyway, in the past, he was subjected to reductive mastectomy for gynecomasty. In that occasion also hypothyroidism was diagnosed, and he was consequently treated with substitutive hormone therapy (sodium levothyroxine, 50 μg/day).

After echo scanning, he underwent an angio computed tomography scan of the supra-aortic trunks that confirmed the presence of an IJV nonocclusive intralumen neoformation with an oval, moderately irregular, and uneven morphology. The lesion was stuck on the posterior–medial face of the mentioned vein, and its dimensions were about 10 × 8.7 × 7.6 mm.

The first diagnosis was controversial between a partial thrombosis of the vein and a primitive endoluminal neoplasia, as it was reported for previous cases.[4] The subsequent Echo-Duplex scan investigation of the supra-aortic trunks, performed before and after echographic SonoVue® (Bracco, Milano) injection, revealed the presence of a markedly hypoechoic, vascularized, and apparently pedunculated neoformation (Fig. 1). The lesion was compressible by the transducer and displayed precocious artery enhancement and progressive washout. This let us to rule out that the mass was ascribable to a clot

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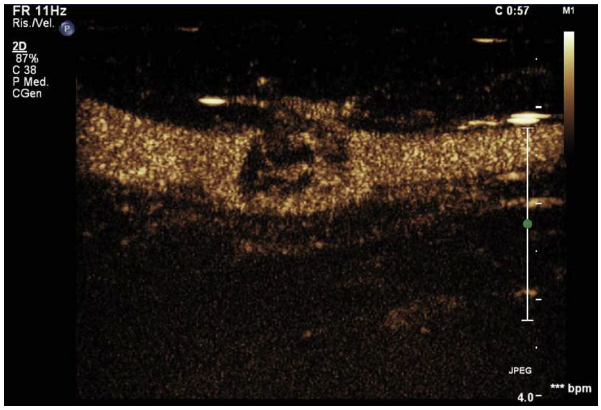

Fig. 1. Echo-Doppler scan investigation after echographic SonoVue injection.

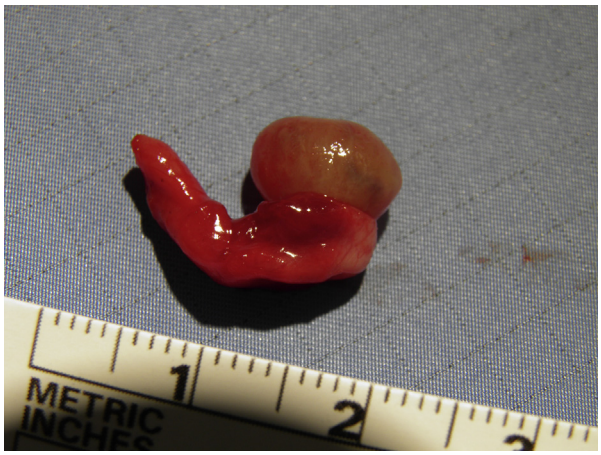

Fig. 2. Excised PG-containing IJV segment.

and suggested a most likely productive process inside the vein.

The neoformation was surgically removed. We approached the IJV by a left supraclavicular incision and then we excised the corresponding vein segment that displayed the internal palpable mass. The proximal and distal stumps were ligated, we didn't perform the IJV reconstruction because of the patency and the good size of the external jugular vein (EJV; Fig. 2). Histologic examination, immunohistochemical coloring for CD-31 (Fig. 3), CD-34, and podoplanin, and histochemical coloring for Alcian-blue pas and Azan Mallory revealed that the neoformation was made up by myxoid stroma and little vascular structures, similar to capillaries.

The morphologic and immunohistochemical scenarios, according to the instrumental evidences mentioned previously, appear to be coherent with the diagnosis of a pyogenic capillary hemangioma (PG).

Finally, 1 month after discharge, the patient underwent an Echo scan which revealed good patency of the EJV.

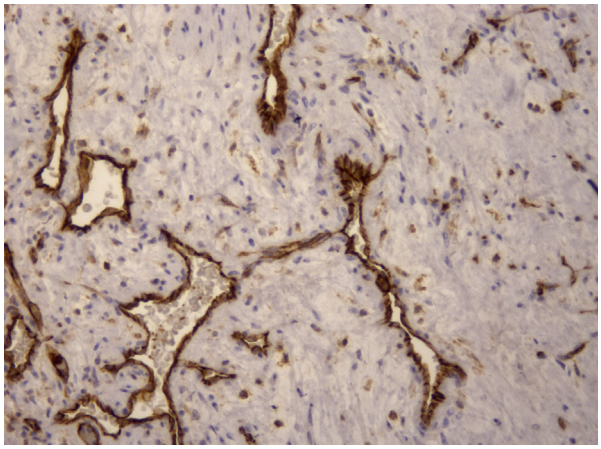

Fig. 3. CD 31 stain outlining vessels' distribution in neoformation (×20 magnification).

DISCUSSION

Literature data concerning PGs localized on the internal walls of the veins are extremely poor, but this fact, almost certainly, does not reflect the real incidence of this pathology. In fact, they are very often asymptomatic and incidentally discovered.

The first documented case of PG is dated back to 1967, when a neoformation in the EJV was detected in a 32-year-old man and a thrombotic event was ruled out after a phlebography.[8]

The improvement of the imaging techniques has later allowed to reach more precise diagnostic pictures. Anyway, their confirmation always needs a surgical approach followed by histologic, histochemical, and immunohistochemical evaluations.

The etiology of these events remains obscure. Some authors describe the existence of congenital venous hemangiomas,[9] but these lesions are detectable also after traumas or infectious events that trigger vascular hyperproliferation. In addition, pressure or hormone shocks or disorders can be related with the incoming of a PG.[10]

Hemangiomas are generally characterized by veins that display a single-layer endothelium and a normal endothelial cell cycle.[3] The peculiar histologic traits of PG and vascular hemangiomas in general, allow to differentiate them from malignant intravascular tumors.[4,11] Thus, a multidiscipline, radiologic, surgical, histologic approach is essential to reach a correct and detailed diagnosis.

To our knowledge, against a certain amount of EJV hemangioma cases reported in the literature, only a couple of cases involving IJV are detectable. They were both correctly characterized only after surgical excision and histologic or immunohistologic analyses, although they displayed a radiologic

aspect that was definitely close to the known cases ascribable to EJV hemangioma.[7,10]

In our case, the patient suffered from a preceding hypothyroidism and the PG presence in the IJV could be a consequence of an associated hormone disorder. The investigations focused onto the control and improvement of the thyroid pathology let us to highlight by chance the intravenous pyogenic neoformation.

Its dimensions, pedunculated shape, and hypoechoic nature, suggested us to deal with a nonthrombotic mass, and to drive the diagnosis toward a primitive productive neoplastic process. This was the main reason which led us to follow a surgical approach.

So, echo-contrast investigation seems to be the gold standard technique for a first-level differential diagnosis as it has also been reported for a recent case.[12] In fact, the diagnostic enhancement becomes more relevant and intense in contrastography. Furthermore, the exposition of patients to ionizing radiation and incoming artifacts ascribable to denture amalgams can be reduced in respect to more invasive ionizing techniques. That is, the results would be devoid of misleading interferences and, moreover, the relative costs of the analysis could be reduced.

Finally, the subsequent postsurgical immunohistochemical investigations confirmed the presence of PG.

In conclusion, although the continuous improvement of radiologic techniques and clinical investigations that allow to perform a correct differential diagnosis of PG from a partial venous thrombosis, endovenous neoformations, such as intravascular leiomyosarcoma or primary lymphoma, must be surgically excised to allow a certain diagnosis. Although in most cases vein's reconstruction is not strictly mandatory, the venous flow continuity restoration may be requested only for some veins, as for example is for the inferior vena cava, to avoid a consequent undesired hypertension.

Thus, the possibility to reach a right diagnosis of IJV PG remains an open challenge because only after surgical excision a definitive diagnosis is possible.

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