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Case report

Epithelioid hemangioma of the internal carotid artery: A case report supporting the reactive pathogenesis hypothesis of this vascular tumor

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ABSTRACT

The pathogenesis of epithelioid hemangioma is intriguing and has remained a controversial topic in the medical literature. Whether its etiology follows a neoplastic or reactive process is unclear, but a history of traumatic insult to the involved region is common. We report a case of epithelioid hemangioma of the internal carotid artery in a child who had undergone cannulization of the internal jugular vein as an infant to receive extracorporeal membrane oxygenation.

1. Introduction

Epithelioid hemangioma, also known as angioblastic lymphoid (or angiolymphoid) hyperplasia with eosinophilia, is an uncommon benign vascular lesion. This vascular tumor usually develops in the head and neck region and presents as cutaneous or subcutaneous reddish papules or nodules. The subcutaneous layer of the face, and the scalp in particular, have been most commonly cited; however, occurrence within the oral cavity, bone, salivary glands, and large vessels has also been reported [1–3]. Large vessel involvement is rare, and a literature search did not reveal any cases involving the internal carotid artery.

Several case reports have linked the onset of this tumor to a history of local trauma. Sun et al. [4] concluded that a hypoxic stimulus triggers the proliferation of endothelial cells via the release of vascular endothelial growth factor, hypoxia-inducible factors, and inflammatory cells such as eosinophils and mast cells. Another case report linked the development of epithelioid hemangioma to an electric welding rod burn [5]. We present the case of a 9-year-old girl with an epithelioid hemangioma obliterating the internal carotid artery and internal jugular vein. We believe she developed this mass as a result of repeated cannulization of a neighboring vessel, the internal jugular vein, during venovenous extracorporeal membrane oxygenation (ECMO) therapy.

2. Case presentation

A 9-year-old girl presented to our pediatric otolaryngology clinic with a progressively enlarging mass on the right side of the neck. Her medical history was significant for primary pulmonary hypertension as an infant, which was managed with ECMO. The patient's chief complaint was dysphagia and a review of systems was negative for fever, chills, tenderness at the site, shortness of breath, and weight loss.

Physical examination revealed a healthy-appearing girl in no apparent distress. Head and neck examination revealed a right-sided 3 cm level III/IV neck mass that was firm, non-tender, and non-pulsatile. No superficial erythema or palpable fluctuance was noted. The mass did not displace with swallowing and was well circumscribed on palpation. Mild tracheal deviation to the left was noted on physical examination; however, she had no signs of airway compromise. Extensive scarring on the upper right side of

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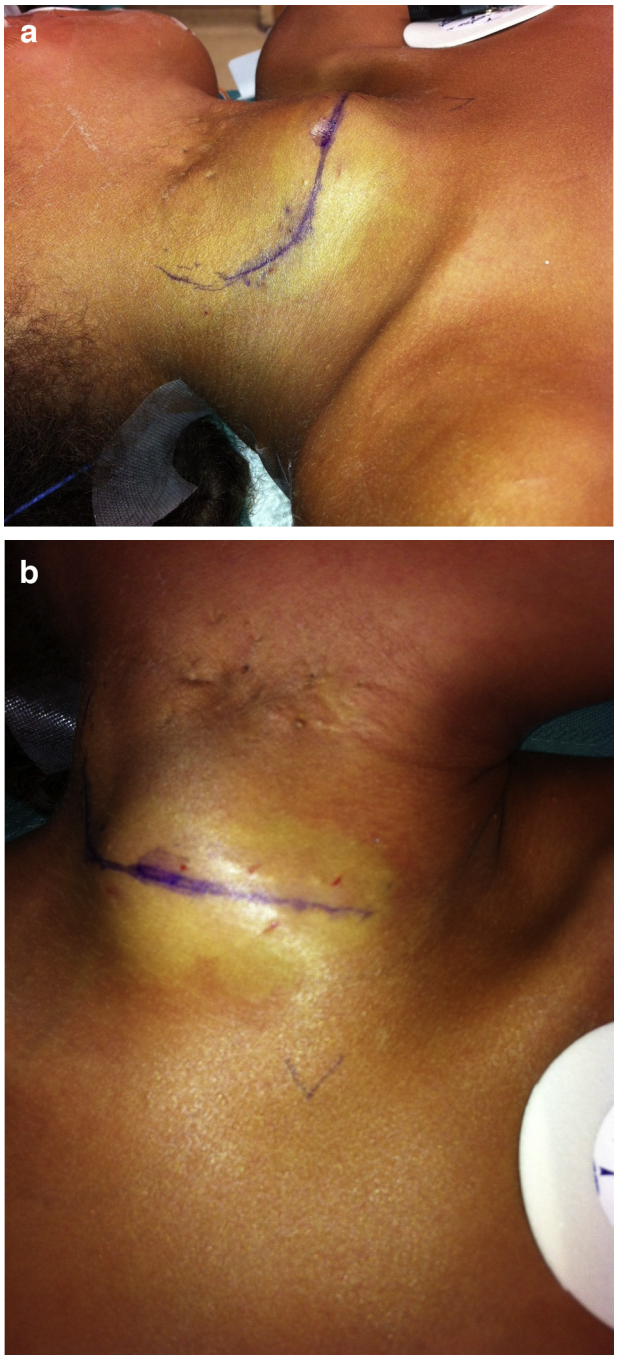

Fig. 1. Lateral (a) and anterior (b) operative photographs taken after injection of local anesthesia demonstrating level III/IV right-sided neck mass and the planned incision line. Note the scars on the right aspect of the neck at the level of the thyroid cartilage consistent with cannulization of the right internal jugular vein for ECMO.

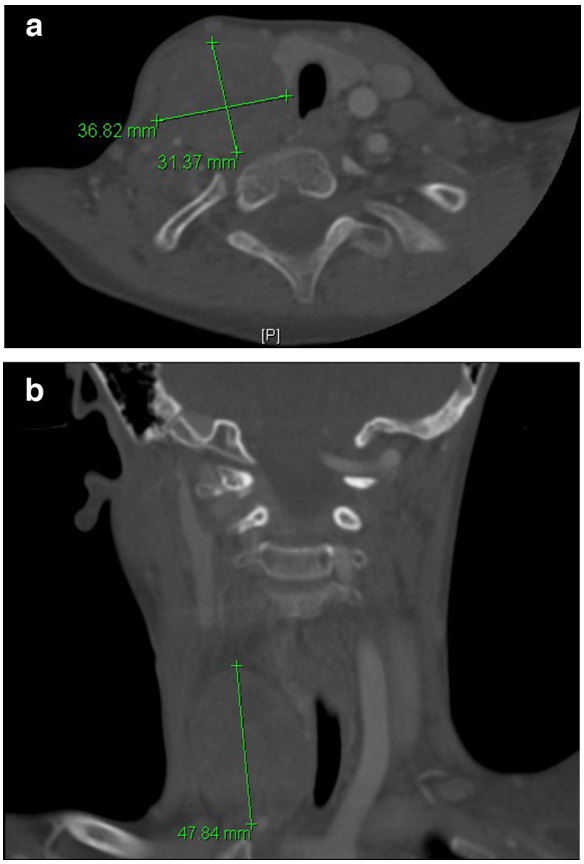

Fig. 2. Axial computed tomography image showing a 4.7 × 3.6 × 3.4 cm right-sided level III/IV mass obliterating a segment of the internal carotid artery as well as the internal jugular vein and compressing the airway (a). Coronal computed tomography image showing the internal carotid artery superiorly and its obliteration inferiorly by the mass (b).

neck suggested that ECMO-related cannulization of the internal jugular vein may have been performed several times throughout the course of her disease as an infant (Fig. 1).

Ultrasound revealed a well-circumscribed heterogeneous mass that was superficial to the right thyroid lobe and deeper to the strap muscles. Computed tomography with contrast enhancement showed a 4.8 × 3.7 × 3.1 cm mass with numerous collateral blood vessels that originated from the right common carotid artery (Fig. 2a). The internal carotid artery and the internal jugular vein appeared to be obliterated by the vascular mass (Fig. 2b).

Preoperative planning included angiography and embolization of feeding vessels to the mass, as well as an occlusion study of the right internal carotid artery. These studies were done the day before her scheduled surgery. Collateral flow to the right carotid system was visualized via patent anterior and posterior communicating arteries. A distinct common carotid artery was not visualized during angiography, and significant mass effect on the trachea was noted. Given the compression of the trachea and the planned surgical excision on the following day, the decision was made to keep the patient intubated for airway protection (Fig. 2b).

The patient was brought to the operating department the day after embolization and underwent local excision of the mass with nerve monitoring. Intraoperatively, the internal carotid artery and the internal jugular vein were not identified. Several feeding vessels to the mass were tied and ligated. The mass was peeled off surrounding structures with minimal blood loss and excised completely. Cranial nerve X was identified and preserved during the excision.

Extubation was performed safely in the operating room after the procedure. Her postoperative course was uneventful. The surgical drain was pulled on day 1 and the patient was sent home on postoperative day 2. After 6 months of follow-up she has been free of recurrence. Histologic analysis under low magnification revealed benign vascular proliferation consisting of well-formed capillary-sized vessels with chronic inflammatory cells favoring.