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Case Report

Spontaneous hematoma caused by arteriovenous malformation of the hyoid bone: A case report

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ABSTRACT

Arteriovenous malformations are congenital vascular anomalies in which there is a direct communication between arteries and veins. Extracranial lesions in the head and neck are rare, and may not be detected until adolescence. We present a case of a 14-year-old male who presented with an expanding neck hematoma. After securing the airway, angioembolization was performed, followed by hematoma evacuation and surgical resection of the lesion. Imaging and histologic examination demonstrated an arteriovenous malformation (AVM) of the hyoid bone. While uncommon, AVMs must be considered in the evaluation of a pediatric neck mass, given the propensity for rapid deterioration.

1. Introduction

Arteriovenous malformations (AVMs) are uncommon vascular anomalies in which a direct communication from artery to vein is present, with no intermediate capillary network [1]. While the majority of these malformations occur intracranially, extradural AVMs of the head and neck are infrequently encountered. Unlike vascular tumors of the head and neck, arteriovenous malformations do not progress in a predictable manner, which makes monitoring and treating these lesions particularly challenging [2].

While the precise underlying mechanism is unclear, arteriovenous malformations are known to arise from abnormal gestational vascular development and morphogenesis [3]. Channels connecting arteries and veins fail to regress, allowing shunting of blood from high to low pressure vessels. Over time, increased blood flow results in the expansion of the lesion and progression of symptoms.

Presenting symptoms can vary greatly, from a cutaneous blush resembling a ‘port wine stain’, to pain and ulceration. The majority of head and neck arteriovenous malformations occur on the face, and are therefore identified in infancy or early childhood. However, up to 41% do not become evident until puberty or later [2]. Primary malformations of bone are particularly difficult to detect, only to be found as a cause of massive bleeding after a dental extraction. Similarly, arteriovenous malformations involving the deep structures in the neck may not be discovered until they reach an advanced stage.

Neck swelling in a child is a common entity, most often due to an infectious etiology, such as reactive lymphadenopathy, lymphadenitis, or deep neck space abscess. A new, rapidly growing mass is usually inflammatory. However, if a patient lacks the characteristic signs of infection such as fever or leukocytosis, there must be a high index of suspicion for both neoplastic and congenital pathologies [4].

While arteriovenous malformations are rarely encountered in the neck, they are more likely to be identified at an advanced stage, and can progress rapidly. Spontaneous hemorrhage is a feared complication of advanced AVMs, and neck hematomas present an extremely challenging clinical scenario. The anatomic relationships are distorted, there is limited time to act, and the sequelae include airway obstruction, cerebral ischemia, and death [5].

In this article, we present a rare case of extracranial arteriovenous malformation involving the hyoid bone, in which spontaneous bleeding resulted in acute airway obstruction. To the authors' knowledge, this article describes the first reported case of an AVM of the hyoid bone.

2. Case report

An otherwise healthy 14-year-old male presented to the emergency department with a one-day history of an expanding left neck mass. Aside from a single episode of neck swelling four years prior, he noted no recent neck mass, swelling, or trauma.

He reported a recent upper respiratory infection, but at the time of presentation, he was afebrile, and without leukocytosis. Physical exam was remarkable for ill-defined swelling of the left neck, which was firm

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and mildly tender to palpation. Initial computerized tomography (CT) scan demonstrated a 0.6 cm × 1.3 cm expansile lesion of the left hyoid bone with an associated large soft tissue mass measuring 4.1 cm × 3.9 cm x 11.2 cm. Due to the poor quality of the images, determining the etiology of the mass based on its radiographic appearance proved difficult. The differential diagnosis based the initial CT included atypical infection, and neoplasm such as rhabdomyosarcoma, lymphoma, or primary bone lesion. Although the CT scan demonstrated rightward shift of the pharynx, larynx, and trachea, initial fiberoptic laryngoscopy revealed normal anatomy without edema or narrowing.

As the initial findings were inconclusive, but concerning for a neoplastic or infectious process, the patient was admitted for further workup, and started on broad-spectrum intravenous (IV) antibiotics. Shortly after admission, however, the patient developed acute dyspnea, immediately following an episode of forceful coughing. He was found obtunded and hypoxic. Orotracheal intubation was attempted, but diffuse edema of the supraglottic larynx obscured the glottic opening, so a laryngeal mask airway was placed. During transport to the operating room for emergent tracheotomy, he maintained normal oxygen saturation and peripheral pulses. However, in the OR, the patient coded three times, requiring a total of four doses of epinephrine and one dose of atropine. As the tracheotomy was performed, copious frothy pink secretions emerged from the trachea, consistent with post-obstructive pulmonary edema. He remained on an epinephrine drip post-operatively, and required aggressive resuscitation with crystalloid and blood products.

After the patient was stabilized, fine needle aspiration of the neck mass was performed at bedside, which showed red blood cells and no evidence of malignancy. Repeat imaging of the neck was also obtained, given both the poor initial study, and the concern for possible hematoma. The new CT of the neck re-demonstrated the expansile hyoid lesion, and better characterized the adjacent mass, previously concerning for neoplasm, as a hematoma with surrounding edema (Fig. 1). The computerized tomographic angiogram (CTA) showed multiple small branches from the left external carotid extending into the region of the lesion, likely including the lingual and facial arteries (Fig. 2). The following day, formal angiogram was performed, which revealed a 3 mm × 2 mm x 2 mm pseudoaneurysm of the proximal segment of the left lingual artery (Fig. 3). Additional findings included moderate hyperemia and neovascularity from multiple small branches of the proximal left external carotid artery. The lingual artery was occluded with coil embolization at the area of the pseudoaneurysm.

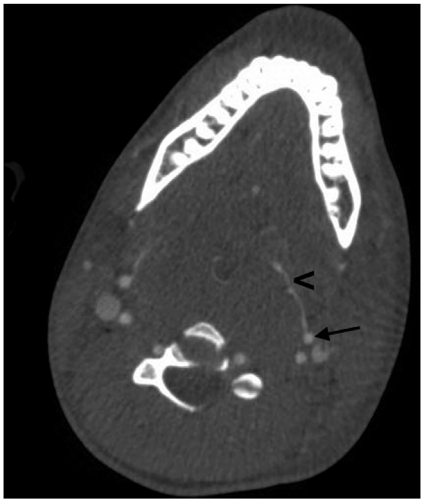

Fig. 2. CT angiogram showing branches (chevron) of the external carotid artery (arrow) extending into the area of the hyoid lesion.

Over the next several days, the patient's status improved. He was weaned off vasopressors, sedation and mechanical ventilation. A complete neurological evaluation revealed no neurocognitive deficits. A magnetic resonance imaging (MRI) study of the neck was obtained to better characterize the lesion prior to resection. The hematoma demonstrated heterogenous T1 shortening, and measured 7.8 cm × 4.1 cm x 3.8 cm, positioned anterior to the left carotid vessels and internal jugular vein, extending into the retropharyngeal and parapharyngeal spaces (Fig. 4). The vascular malformation was not well visualized on MRI, due to the large surrounding hematoma and motion artifact.

On hospital day ten, the patient was taken for evacuation of the hematoma and resection of the vascular malformation. Intraoperatively, a large amount of clotted blood was encountered, along with small fragments of bone. The left hyoid body and superior cornu were largely absent, replaced by a soft tissue mass and dilated bony cyst. Several small vessels were seen extending into this area and were ligated. Bony fragments, soft tissue, and adjacent normal hyoid bone were sent for histologic examination. The pathologic findings included large and small abnormal vessels, benign appearing spindle cell proliferation, and a secondary aneurysmal bone cyst within the hyoid

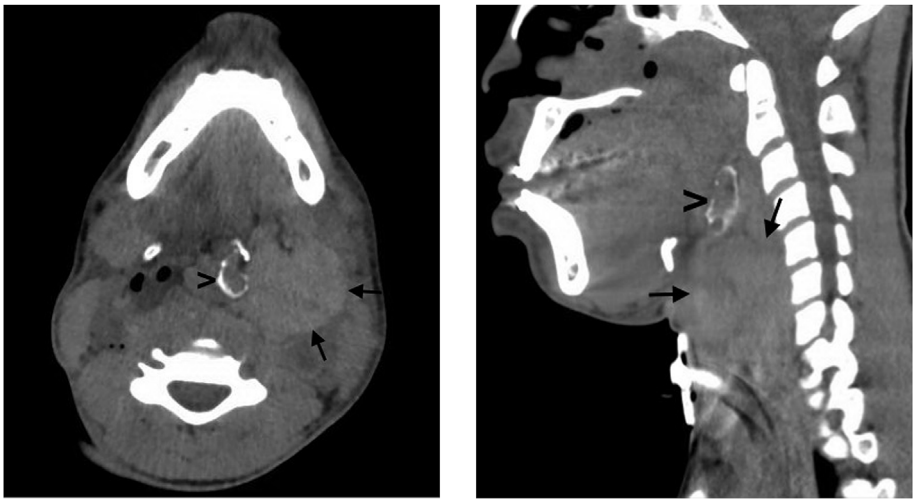

Fig. 1. (a): Axial computerized tomography (CT) scan without contrast demonstrating the hematoma (arrows) and cystic hyoid bone (chevron). (b): Sagittal CT without contrast demonstrating the hematoma (arrows) and cystic hyoid bone (chevron).