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Diffuse infiltrative laryngoesophageal and peritoneal venous malformations mimicking carcinomatosis with a subclavian vein aneurysm☆

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ABSTRACT

Venous malformations are benign vascular lesions that can occur in any part of the body. Esophageal venous malformations can extend into the peritoneum, so preoperative imaging can be decisive in diagnosis and treatment. The characteristic manifestations of venous malformations are transmural phleboliths, centripetal enhancement on dynamic CT, and bluish mucosa on endoscopy. We report a rare case of female patient diagnosed with venous malformations involving laryngoesophagus, mediastinum, and peritoneum, which mimicked carcinomatosis, in addition to a left subclavian vein aneurysm.

1. Introduction

According to International Society for the Study of Vascular Anomalies, vascular lesions are classified into two categories: hemangiomas representing endothelial tumor and congenital vascular malformation. However, the term "hemangioma" has been used incorrectly to describe all the vascular anomalies, regardless of their pathogenesis [1]. According to its definition, the term "hemangioma" should represent only vascular tumor. It must be pointed out that what has been called "cavernous hemangiomas" are virtually "venous malformation," histologically composed of dilated vascular channels resulting from defect in vascular smooth muscle recruitment in vascular morphogenesis [2,3].

Vascular anomalies of the gastrointestinal (GI) tract such as venous malformations are rare, benign vascular tumors most commonly found in the small intestine [3,4]. The esophagus is the least common site for GI venous malformations, accounting for only 3.3% of all benign esophageal tumors [5]. In general, about 60–80% of all venous malformations are found in the head and neck region. Among those, laryngeal venous malformations are rare, especially in adults [6]. There have only been a few case reports dealing with adult neck venous malformations, which were mostly located in the supraglottic area. It has been reported that large neck venous malformations in adults can extend into the mediastinum and rupture spontaneously, resulting in mediastinal fluid collection [7]. However, no previous diffuse esophageal venous malformations accompanied by multifocal satellite lesions extending to the vocal cords, lower cervical neck, mediastinum, and peritoneum have been reported. In this report, we describe an unusual case of diffuse infiltrative esophageal venous malformation concomitant with left subclavian vein aneurysm. The purpose of this report is to elucidate sonographic, gastroscopic, and computed tomography (CT) findings of diffuse infiltrative venous malformation, allowing for the final diagnosis to be assessed preoperatively.

2. Case report

A 54-year-old female patient was admitted to our hospital for thyroid cancer surgery. After admission, she underwent neck ultrasound (US) and contrast-enhanced neck CT for preoperative evaluation of thyroid cancer and endoscopy for evaluation of dysphagia. The US incidentally showed a marked 1.3-cm hypoechoic mass in the left vocal cord (Fig. 1a) in addition to the thyroid malignancy. Neck CT showed the left vocal cord mass to have heterogeneous enhancement (Fig. 1b). In addition, multiple enhancing soft tissue masses were incidentally detected at both levels VI and VII of the neck.

In order to further evaluate the enhancing soft tissue masses found in the lower neck area, we analyzed a contrast-enhanced chest CT and multiphase abdominal CT. These images showed three additional irregular enhancing lesions in the mediastinum. In addition, we observed extensive esophageal wall thickening from the proximal esophagus to the esophagogastric junction on the CT. There were multiple calcific foci within the thickened esophageal wall (Fig. 2). The endoscopic exam showed dilated varix-like vessels with bluish overlying mucosa (Fig. 3), concordant with esophageal varices, which are frequently found in advanced liver cirrhosis. However, there were no evidence of

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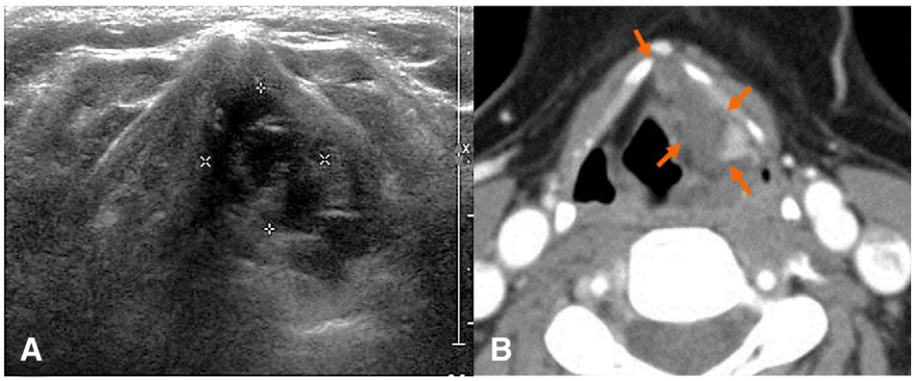

Fig. 1. (A) Neck US shows a 1.3-cm hypoechoic mass in the left vocal cord. (B) Neck CT shows a heterogeneous enhancing soft tissue mass in the left vocal cord, obliterating the fat plane.

liver cirrhosis and varix on multiphase abdominal CT. Incidentally, we found additional multiple enhancing soft tissue masses adjacent to the distal greater curvature of the stomach and along the peritoneum (Fig. 4a–c). Those soft tissue masses showed peripheral nodular enhancement and progressive centripetal fill-in, similar to liver hemangiomas. Moreover, a focal outpouching sac at the inferior aspect of the left subclavian vein was incidentally detected (Fig. 5). We assessed the coronal, sagittal, and three-dimensional (3D) reconstruction images through a reconstruction program (Aquarius iNtuition, TeraRecon, Inc) and concluded that the outpouching sac was consistent with saccular aneurysmal dilatation of the subclavian vein.

To our knowledge, however, there has been no report presenting diffuse esophageal venous malformations accompanied by multifocal satellite lesions extending to the vocal cords, lower cervical neck, mediastinum, and peritoneum that we performed positron emission tomography (PET) CT scan for the exclusion of mixed malignancy or metastasis. PET-CT scan showed no abnormal uptake in the multifocal enhancing soft tissue lesions extending from the vocal cords to the peritoneum as well as in the diffusely thickened esophageal wall. Finally, we were able to preoperatively diagnose a diffuse infiltrative laryngoesophageal and peritoneal venous malformations rather than carcinomatosis.

Consequently, the patient underwent total thyroidectomy with lymph node dissection for known primary thyroid cancer, which was confirmed to be papillary thyroid cancer. Additional biopsy of the paratracheal soft tissue mass was also collected during surgery. The histopathological findings showed irregularly dilated thin-walled vascular channels with some fibroadipose tissues, suggesting venous malformation (Fig. 6).

Because the imaging findings from various imaging modality including US, CT, endoscopy, and PET-CT scan were consistent with a low-flow venous malformation, there was no need for further surgical biopsy for the other soft tissues lesions in order to exclude primary thyroid cancer metastasis or other malignancy. The patient was prescribed regular 6-month follow-ups and had been no symptom or sign of disease progression. Furthermore, some of the peritoneal lesions showed no interval change in the size and shape on follow-up CT image 2 years later

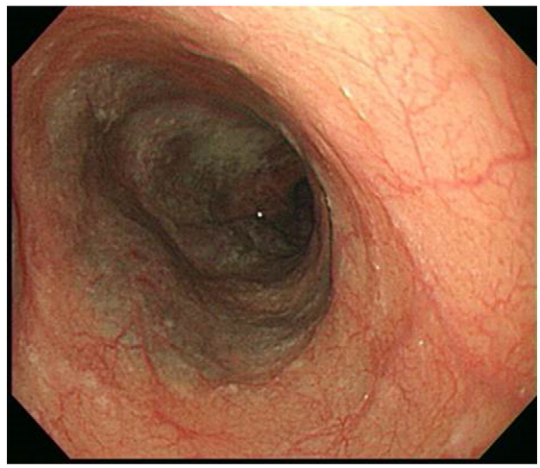

Fig. 3. Gastroendoscopic exam shows a bluish tubular submucosal lesion similar to an esophageal varix.

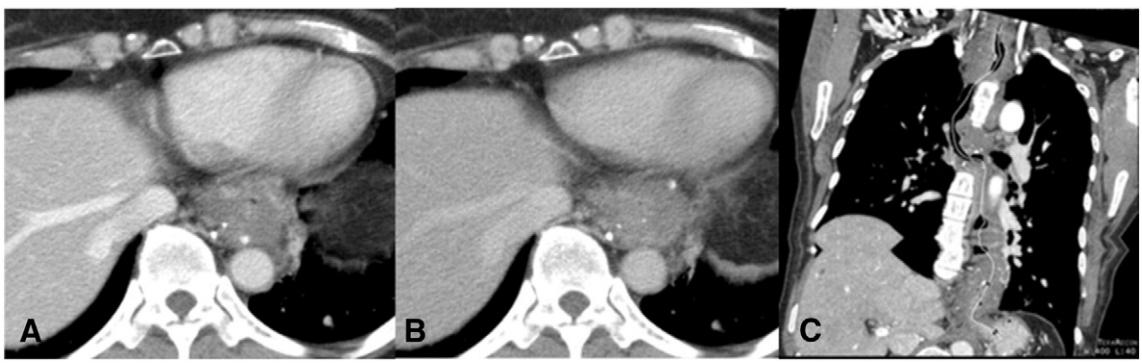

Fig. 2. Portal (A) and 3-min delayed (B) dynamic chest CT images show marked circumferential esophageal wall thickening from the proximal esophagus to the esophagogastric junction. Multiple calcific foci in the thickened esophageal wall and a centripetal enhancement pattern are also shown in the 3-min delayed phase. (C) Curved coronal multiplanar reconstruction image shows diffuse esophageal wall thickening from proximal to esophagogastric junction with some phleboliths.