

Oesophageal cavernous haemangioma

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Key words

Esophageal haemangioma • Benign esophageal tumors • Benign esophageal neoplasms • Cavernous haemangioma

Summary

Esophageal cavernous haemangioma is an uncommon benign neoplasm. These tumors are usually discovered incidentally as they are often asymptomatic. The symptoms, if present, are bleeding and dysphagia. Endoscopic and radiographic features are non-specific and histopathologic examination is required for definitive diagnosis and appropriate treatment. We herein report a case of a

69-year old man who presented with complain of mild dysphagia for solid foods. Endoscopic evaluation with transesophageal ultrasonography and CT revealed a 5 cm intramural tumor in the posterior wall of the upper esophagus. The tumor was resected and histological examination showed an esophageal cavernous haemangioma.

Introduction

Esophageal cavernous haemangioma is an uncommon neoplasm and comprises less than 5% of esophageal benign tumors ¹. Cavernous haemangiomas are benign neoplasm of mesenchymal origin with vascular proliferation and composed of dilated, thin-walled blood vessels lined by flattened endothelium. These lesions most commonly involve skin, head and neck soft tissues, liver and brain. But they can arise at almost any location. Esophageal cavernous haemangiomas are submucosal lesions. They are usually discovered incidentally as the patients are often asymptomatic ². The symptoms, if present, are bleeding and dysphagia ³. Surgical resection and histological evaluation is necessary to confirm the diagnosis as malignancy has been reported in association with these lesions, especially in tumors larger than 3 cm ^{4,5}. Endoscopic resection is recommended if the diagnosis is certain, the tumor is less than 2.5 cm and in the absence of bleeding ⁶. The tumor is usually asymptomatic but it can cause hemorrhage and, if large enough, dysphagia ⁷. Often histological evaluation is required as the imaging findings and endoscopic appearance of these lesions are nonspecific ⁸. In our case a leiomyomatous tumor was suspected based on the imaging findings.

Presentation of case

A 69-year-old man presented with occasional transient dysphagia for solid foods. An initial upper gastrointestinal endoscopic evaluation with transesophageal ultrasonography (Fig. 1 A-B) revealed a 5 cm polypoid mural lesion along the posterior wall of the upper esophagus with intraluminal protrusion and a pseudopedunculated appearance, stretching the blood vessels. A subsequent CT scan confirmed the presence and location of the lesion and showed thickening of the distal esophageal wall (Fig. 2 A-B). The entire lesion was resected by video-assisted thoracotomic surgery (Fig. 3 A-B) with an unremarkable postoperative course and no complications. The gross pathological examination revealed a 4 x 5 cm specimen. The soft surface of the specimen was blue white to blue purple (Fig. 4 A). After fixation with formalin for 24 hours, the sections through the mass showed spongy hemorrhagic tissue with an apparent nodular component (Fig. 4 B). The microscopic evaluation showed a circumscribed lesion that was composed of irregular dilated thin-walled vessels lined by flattened single layer of endothelial cells without evidence of any malignant cells, a thick muscle coat or an internal elastic lamina (Fig. 5 A-B). Immunostaining with Desmin, MSA (HHF35), CD34, CD31

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Fig. 1. Endoscopic finding of the huge protruding tumor in the upper esophagus.



Fig. 4. A) The resected specimen fresh measuring 50X40 mm; B) The specimen after 24 hour of formalin fixation gets a more clear nodular aspect.

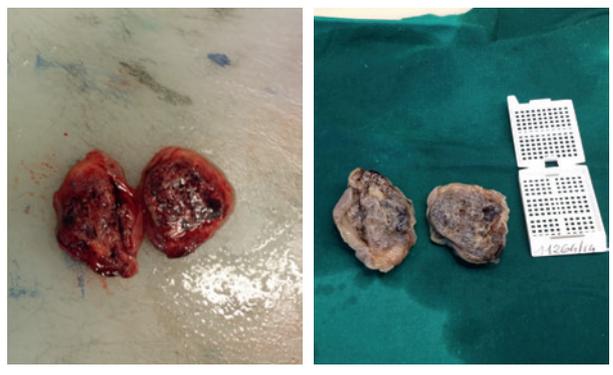


Fig. 5. A) Histological examination of the resected specimen shows the dilated irregular venous channels with the size larger than that of capillaries in the submucosa; H.E: original magnification X20; B) The elastic stain demonstrates the absence of internal elastic lamina in the large vessels; original magnification X40.

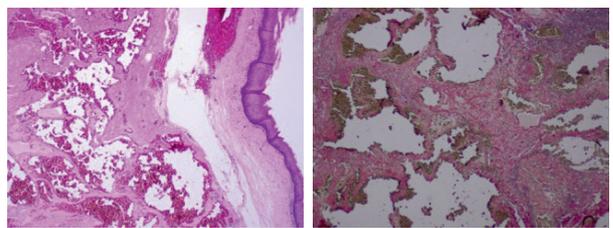


Fig. 2. (A-B) Chest CT scan reveals a polypoid tumor of the posterior wall of the upper portion of esophagus.

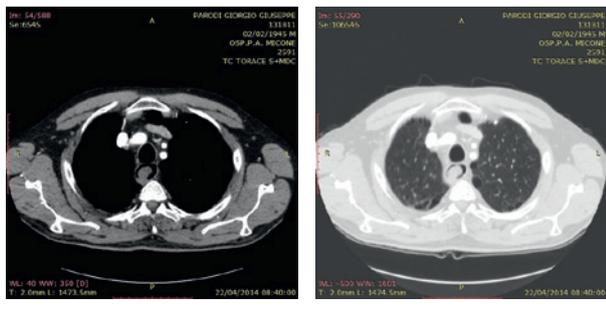
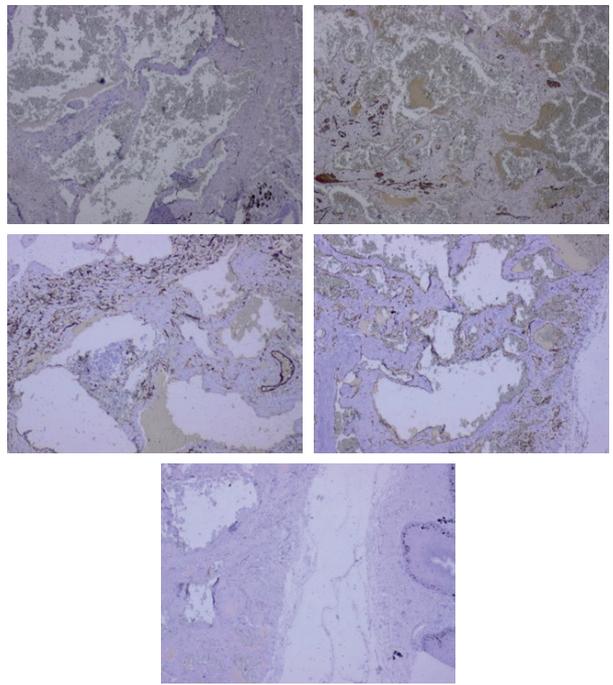


Fig. 3. (A-B) Intraoperative images show the large polypoid lesion of the posterior wall of upper esophagus, ligated at the base before complete resection.



Fig. 6. A-B) Immunostain for Desmin (A) and MSA (HHF35): X40 (B) demonstrate the absence of muscle coat in the vascular walls; C-D)- Immunostain for CD34 and CD31 highlights a monolayer of endothelial cells without any malignancy: X40; E)- Growth fraction. Staining for Ki67/MIB1: X20 Original magnification: A, B, C, X40; D, EX20



and inconspicuous proliferation index (Fig. 6 A-B-C-D-E) confirmed the diagnosis of cavernous haemangioma.

Discussion

Esophageal haemangiomas are relatively rare and are positioned in submucosa⁸. Haemangiomas are benign vascular proliferations and classified histologically into cavernous, capillary, hamartomatous and arteriovenous subtypes. The cavernous haemangioma is defined by the size of the venous channels that are larger than capillaries⁸. These tumors should be differentiated from malignant haemangiomas, which are classified histologically into haemangioendothelioma, endoscopic findings have been described as a blue colored protruding submucosal tumor. The lesion can be seen anywhere in the esophagus and can have atypical appearance such as reddish discoloration, a color similar to the normal mucosa and ulceration. In our case the lesion had a color similar to the normal mucosa. The endoscopic and radiologic appearance of the lesion did not establish the diagnosis of a haemangioma. We performed both histochemical and immunohistochemical stains to confirm the diagnosis and differentiate the lesion from arteriovenous malformation and intramuscular haemangioma.

Conclusion

Esophageal haemangiomas are rare and often small benign submucosal neoplasms. In our case the lesion was large with thickening of the distal esophageal wall and nonspecific imaging and endoscopic features. Definitive diagnosis was possible only by histopathological examination.

References

- ¹ Moersh HJ, Harrington SW. *Benign tumour of the esophagus*. Ann Otol Rhinol Laryngol 1944;53:800-17.
- ² Platcha A. *Benign tumours of the esophagus; review of literature and report on 99 cases*. Am J Gastroenterol 1962;38:639-52.
- ³ Plachta A, Schmidt HW. *Benign tumours and cysts of the esophagus*. J Thorac Cardiovasc Surg 1961;41:717-32.
- ⁴ Eda Y, Asaki S, Yamagat L, Ohara S, et al. *Endoscopic treatment for submucosal tumours of the esophagus: studies in 25 patients*. Gastroenterol Jpn 1990;25:411-6.
- ⁵ Bandoh T, Isoyama T, Toshima H. *Submucosal tumours of the stomach. A study of 100 operative cases*. Surgery 1993;113:498-506.
- ⁶ Yoshikane H, Suzuki T, Yoshioka N, et al. *Hemangioma of the esophagus: endosonographic imaging and endoscopic resection*. Endoscopy 1995;27:267-9.
- ⁷ Tominaga K, Arakawa T, Ando K, et al. *Oesophageal cavernous haemangioma diagnosed histologically, not by endoscopic procedures*. J Gastroenterol Hepatol 2000;15:215-9.
- ⁸ Chella B, Nosotti M, Baisi A, et al. *Unusual presentation of a trans-paraesophageal cavernous hemangioma of the esophagus*. Dis Esophagus 2005;18:349-54.