Jikeikai Med J 2014 ; 61 : 53-7

Case Report

A Huge Esophageal Hamartoma Treated with Endoscopic Resection

Toshiyuki SASAKI[1], Nobuo OMURA[1], Yuichiro TANISHIMA[1], Katsunori NISHIKAWA[1], Norio MITSUMORI[1], Hideyuki KASHIWAGI[1], Hirobumi TOYOIZUMI[2], and Katsuhiko YANAGA[1]

[1]*Departments of Surgery, The Jikei University School of Medicine*
[2]*Department of Endoscopy, The Jikei University School of Medicine*

ABSTRACT

We report a huge hamartoma that developed in a 44 years old man who presented to our clinic with dysphagia. Upper gastrointestinal endoscopy demonstrated a giant pedunculated polyp that protruded from the esophageal entrance. Esophagography revealed a smooth-surfaced mass measuring 13×4×3 cm with a stalk measuring approximately 2×2 cm. These findings, together with those of computed tomography and magnetic resonance imaging of the chest, led to a tentative diagnosis of fibrovascular polyp. The patient underwent endoscopic resection with a snare under general anesthesia, and the tumor was successfully excised with segmental resection. The final pathological diagnosis was hamartoma of the esophagus. The patient had a favorable postoperative course and was discharged from the hospital in good general condition. He has been followed up on an outpatient basis and, to date, has had no symptoms or evidence of recurrence.

(Jikeikai Med J 2014 ; 61 : 53-7)

Key words : hamartoma, esophagus, esophageal polyp, benign tumor, endoscopic resection

INTRODUCTION

Hamartoma is a benign, focal hyperplasia of normal and mature tissue composed of one or more cellular elements. It develops most frequently in the lungs, heart, or hypothalamus but rarely in the gastrointestinal tract[1]. We report a giant esophageal hamartoma that was resected endoscopically under general anesthesia.

CASE PRESENTATION

A 44 years old man had first experienced a feeling of food getting stuck in his throat upon swallowing approximately 20 years ago, but did not seek medical care at the time. In July 2008, he went to a nearby hospital with a chief complaint of dysphagia and worsening chest discomfort. Because computed tomography (CT) of the chest indicated a possible mass lesion in the esophagus, the patient was referred to our hospital for further evaluation and treatment in October 2008.

On physical examination, the patient was 175 cm tall and weighed 60 kg. There was no conjunctival pallor or jaundice, and no mass was palpable in the neck. There was no rash on the face or upper extremities and no papilloma in the mouth characteristic of Cowden's disease. No enlarged cervical lymph nodes were found, and the thyroid gland was of normal size. There was no jugular vein distention. The results of laboratory studies were normal.

Upper gastrointestinal endoscopy showed a smooth-surfaced, pedunculated lesion that protruded from below

Received for publication, July 25, 2014
佐々木敏行，小村　伸朗，谷島雄一郎，西川　勝則，三森　教雄，柏木　秀幸，豊泉　博史，矢永　勝彦
Mailing address : Toshiyuki SASAKI, Department of Surgery, The Jikei University School of Medicine, 3-25-8 Nishi-shimbashi, Minato-ku, Tokyo 105-8461, Japan.
E-mail : sasakit@jikei.ac.jp

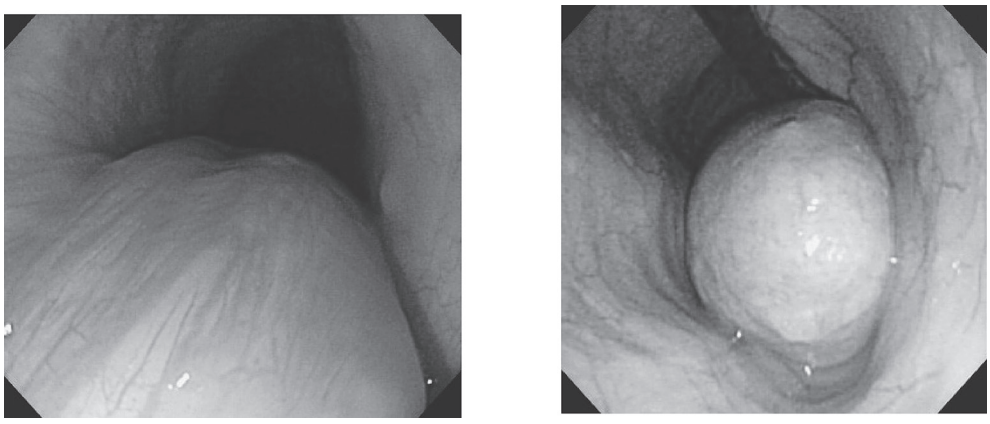

Fig. 1. Upper gastrointestinal endoscopy showed a smooth-surfaced pedunculated lesion that protruded from below the esophageal entrance.

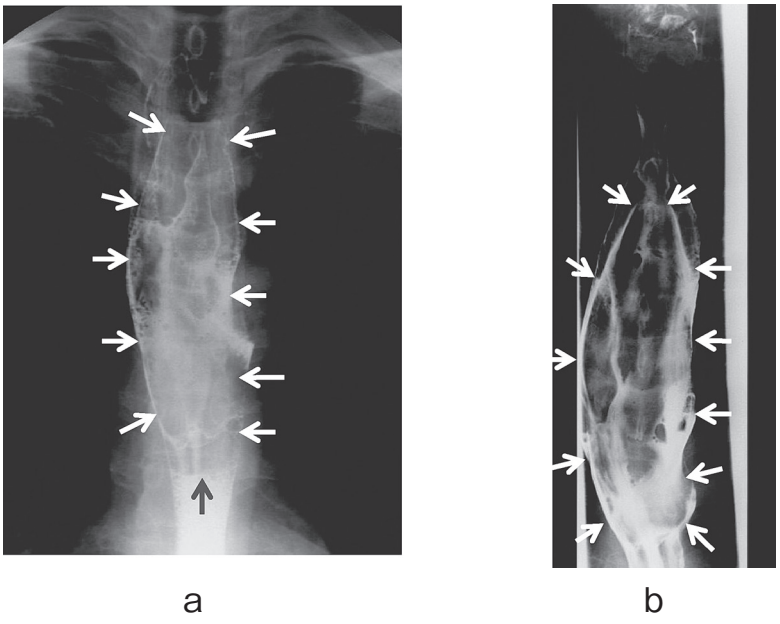

Fig. 2. Esophagography showed a 13×4×3-cm mass.

the esophageal entrance (Fig. 1a, 1b). No abnormalities were found in the stomach or duodenum. Upper gastrointestinal endoscopic biopsy showed fibrous connective tissue including blood vessels and stratified squamous epithelium with interstitial infiltration. Because of these findings, a fibrovascular polyp was suspected.

Esophagography showed a smooth-surfaced mass measuring 13×4×3 cm with a 2×2-cm stalk (Fig. 2a, 2b). Chest CT revealed a mass with nonhomogeneous partial calcification occupying a region extending from the upper chest to the middle of the esophagus (Fig. 3a, 3b). Magnetic resonance imaging (MRI) of the chest revealed a mass with a well-defined margin containing a cystic component and calcification appearing as a high-intensity area on T1-weighted images and as a low-intensity lesion on T2-weighted images (Fig. 4a, 4b). The findings of CT and MRI were suggestive of a fibrovascular polyp.

Because of the patient's intense feeling of heaviness in the chest during swallowing, resection of the polyp was scheduled. We originally planned open surgery via a cer-

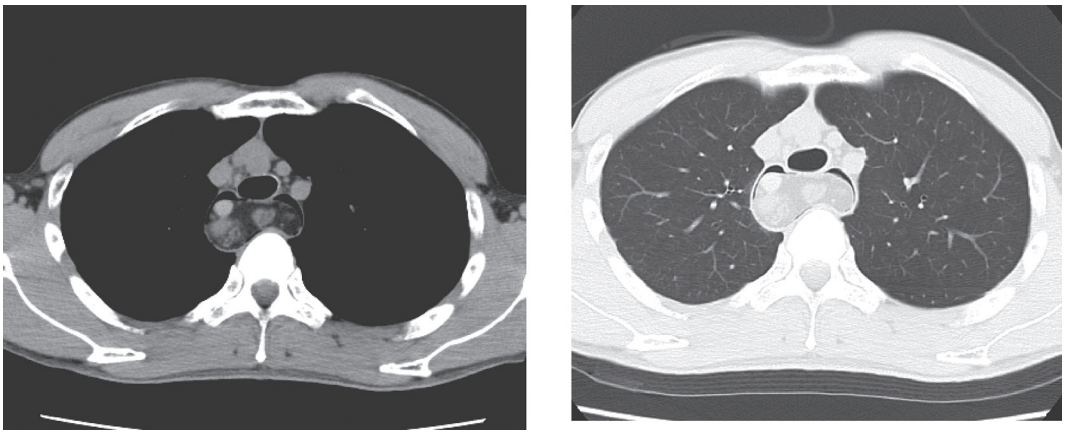

Fig. 3. Computed tomography : a) mediastinal window and b) lung window

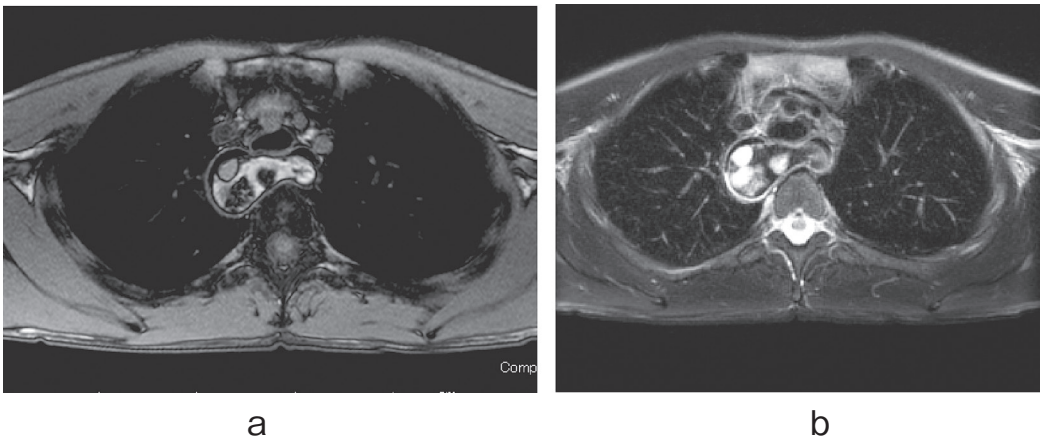

Fig. 4. Magnetic resonance imaging : a) T1-weighted image and b) T2-weighted image

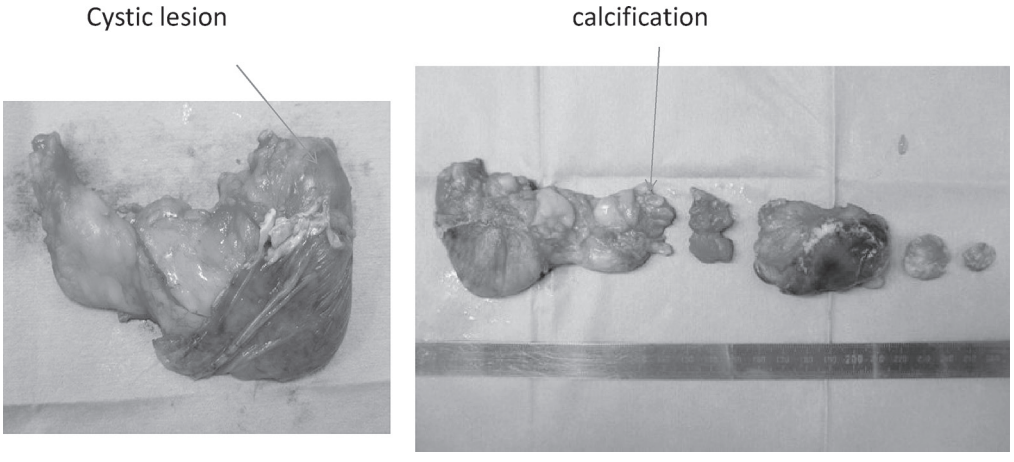

Fig. 5. Resected specimen

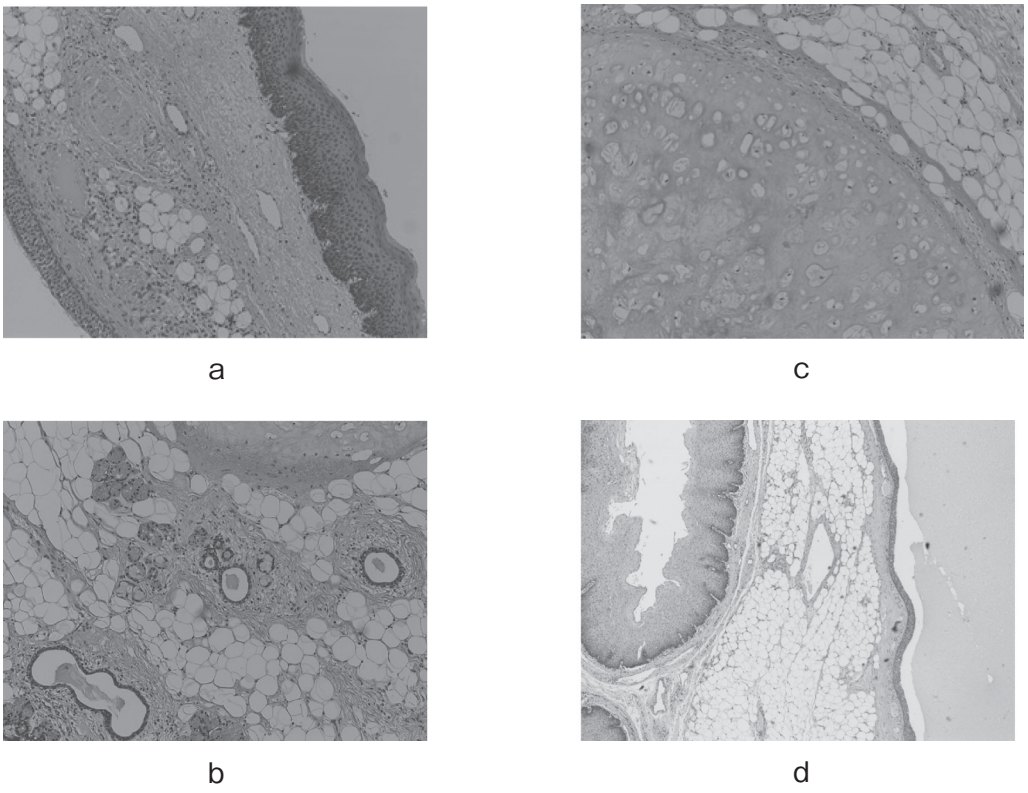

Fig. 6. Pathological examination showed : a) cysts covered with normal esophageal stratified squamous epithelium ; b) iter, mucous glands, and serous glands ; c) bone formation ; and d) cysts covered with normal columnar epithelium (hematoxylin and eosin ×40).

vical approach because the mass was larger than 10 cm. However, following the patient's request, we attempted endoscopic resection under general anesthesia in February 2009. Although *en bloc* resection could not be performed, segmental resection in 5 pieces could be accomplished with a snare. The surgery lasted 1 hour 41 minutes. Blood loss was minimal, and the surgery was completed without complications.

The resected mass measured 13×4 cm ; was composed mainly of soft, yellow, fatty tissue ; and was covered by apparently normal esophageal mucosa (Fig. 5). In addition, some parts of the mass were hard ; therefore, the mass was assumed to be composed of calcified and cystlike regions.

Histopathological examination showed that the lesion was covered with normal esophageal stratified squamous epithelium. No epithelial atypia was noted. The mass was composed mainly of mature fat tissue. Within the mass, morphologically irregular pieces of cartilage were seen, either alone or in groups, and partial bone formation was identified. In addition, cysts covered with normal columnar epithelium and scattered iter, mucous glands, and serous glands were noted (Fig. 6a–d). On the basis of these findings, a definitive diagnosis of hamartoma was established.

Postoperative Course

The patient became capable of oral intake on postoperative day 4 and was discharged on postoperative day 9. The preoperative dysphagia resolved, and the patient could eat without problems. He has been followed up at our outpatient clinic for approximately 2 years since surgery, and no evidence of recurrence or dysphagia has been noted.

Discussion

The most of hamartomas in the gastrointestinal tract occur as a component of Cowden's disease. Solitary hamartomas rarely develop in the esophagus. The most of pri-

Table 1. Reported cases of solitary hamartoma of the esophagus

Author (Reference number)	Year	Age (years)	Sex	Chief complaint	Location	Greatest diameter (cm)	Operative method
Smith et al. (9)	1976	3	F	failure to gain weight	distal	1.5	thoracotomy
Shah et al. (8)	1974	60	M	chest pain	cervical	6	percutaneous transcervical
Beckerman et al. (7)	1980	2	M	failure to gain weight	cervical	2.5	percutaneous transcervical
Saitoh et al. (6)	1990	40	F	hemoptysis	cervical	6	percutaneous transcervical
Lakhkar et al. (5)	1991	30	F	dysphagia	cervical	unknown	percutaneous transcervical
Halfhide et al. (4)	1995	41	M	dysphagia	cervical	12	endoscopic resection
Present case	2011	40	M	dysphagia	cervical	13	endoscopic resection

mary esophageal masses are malignant tumors, mostly cancers, and benign tumors are rare[2]. Benign esophageal tumors include fibrovascular polyps, lipomas, inflammatory polyps, and, most frequently, leiomyomas[3].

A search of the PubMed database with "hamartoma" and "esophagus" as key words yielded 6 cases of solitary hamartoma developing in the esophagus[4–9]. The average patient age in these cases was 30.8 years, and all patients but one had symptoms. In most cases, the tumor developed in the cervical esophagus, and only 1 case was treated with endoscopic excision (Table 1). Histopathologic examination in most previous cases showed cartilage, bone formation, glands, and fat tissue, as was shown in our case.

When a benign mass in the gastrointestinal tract is diagnosed, surgical intervention is generally not indicated unless the patient is symptomatic. In the present case, the mass was large, and the patient complained of dysphagia. Thus, we decided to perform resection. For giant polypoid lesions, surgical resection is usually selected[10,11]. For our patient, however, although the lesion was extremely large, we chose to perform endoscopic resection, because the patient had a strong desire to avoid esophagectomy, the tumor stalk was small (approximately 2 cm), the lesion was considered benign, and an adequate surgical field could be secured as long as the patient was under general anesthesia. *En bloc* resection was not possible, but the mass was successfully removed in 5 pieces.

References

1. Rubin R, Strayer DS, editors. Rubin's pathology : clinicopathologic foundations of medicine 5th ed. Philadelphia : Lippincott Williams & Wilkins ; 2008. p. 227.
2. Attah EB, Hajdu SI. Benign and malignant tumors of the esophagus at autopsy. J Thorac Cardiovasc Surg. 1968 ; 55 : 396–404.
3. Choong CK, Meyers BF. Benign esophageal tumors : introduction, incidence, classification, and clinical features. Semin Thorac Cardiovasc Surg. 2003 ; 15 : 3–8.
4. Halfhide BC, Ginai AZ, Spoelstra HA, Dees J, Vuzevski VD. Case report : a hamartoma presenting as a giant oesophageal polyp. Br J Radiol. 1995 ; 68(805) : 85–8.
5. Lakhkar BN, Ghosh MK, Shenoy PD, Patil UD. Hamartoma : a benign intraluminal tumor of the oesophagus (a case report). J Postgrad Med. 1991 ; 37 : 235–7, 236A–236B.
6. Saitoh Y, Inomata Y, Tadaki N, Mimaki S. Pedunculated intraluminal osteochrondromatous hamartoma of the esophagus. J Otolaryngol. 1990 ; 19 : 339–42.
7. Beckerman RC, Taussig LM, Froede RC, Coulthard SW, Firor H, Tonkin I. Fibromuscular hamartoma of the esophagus in an infant. Am J Dis Child. 1980 ; 134 : 153–5.
8. Shah B, Unger L, Heimlich HJ. Hamartomatous polyp of the esophagus. Arch Surg. 1975 ; 110 : 326–8.
9. Smith CW, Murray GF, Wilcox BR. Intramural esophageal hamartoma : an unusual cause of progressive stricture in a child. J Thorac Cardiovasc Surg. 1976 ; 72 : 315–8.
10. Yannopoulos P, Manes K. Giant fibrovascular polyp of the esophagus—imaging techniques can localize, preoperatively, the origin of the stalk and designate the way of surgical approach : a case report. Cases J. 2009 ; 2 : 6854.
11. Caceres M, Steeb G, Wilks SM, Garrett HE Jr. Large pedunculated polyps originating in the esophagus and hypopharynx. Ann Thorac Surg. 2006 ; 81 : 393–6.