Case Report

Giant Pedunculated Atypical Lipomatous Tumor of Esophagus: A Six-Year Delay Surgical Removal without Adverse Event

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Benign esophageal tumors are rare and usually have no symptoms until enlarging. Most common esophageal polyps are fibrovascular polyps that are difficult to differentiate from atypical lipomatous tumor. A locally aggressive mesenchymal tumor was reported as a large intraluminal polyp because of similar presentation until the pathological examination was done. Usual recommendation is to remove the tumor as soon as possible due to the risk of sudden death. We report on a case where a six-year delay to remove an atypical lipomatous tumor presented with a very large esophageal polyp. Although a lot of cases use endoscopy for resection, longitudinal esophagotomy is a simple and safe procedure for removal of polyps in difficult situations.

Keywords: *Benign esophageal tumor, Atypical lipomatous tumor of esophagus, Giant pedunculated polyp*

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Benign esophageal tumors are rare, as surgical resection are less than 5% in all surgical resections of all esophageal tumors[1]. Fibrovascular polyp is the most common benign tumor of the esophagus, and usually rises from the upper esophagus. The polyps often progress slowly along the esophagus and patients have little or no initial symptoms. Most patients are diagnosed only after the polyp has grown large. The presentation of symptoms is varied, and includes dysphagia, vomiting, retrosternal pain, shortness of breath, and regurgitation of a mass into the oral cavity. Atypical lipomatous tumor is a large intraluminal polyp that presents itself similar to a large fibrovascular polyp, which is difficult to differentiate until pathological examination. The pathology of atypical lipomatous tumors are dissimilar to fibrovascular polyp. There is a locally-aggressive mesenchymal neoplasm composed with potential for local recurrence. Removal of the tumor can be performed by minimally-invasive surgery or open approach with free margin, depending on the location and size of the tumor. Many reports have been published and most tumors were removed at the time of diagnosis. In this report we present a case of large pedunculate atypical lipomatous tumor of the esophagus that was observed for six years after diagnosis without adverse events and successful removal by open approach without complication.

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Case Report

An 80-year-old Thai male had a history of prostate cancer and underwent laparoscopic prostatectomy in 2008. During surveillance for prostate cancer in 2010, we performed computed tomography of his chest and abdomen and found a large esophageal intraluminal polyp arising from the upper esophagus (Fig. 1). The patient was sent for further investigation. The esophagogastroduodenoscopy (EGD) showed a long smooth esophageal polyp beginning at 18 cm from incisors to 30 cm from incisors. The polyp showed no evidence of bleeding. Endoscopic ultrasound showed a homogeneous echo in the polyp and bright areas in the proximal end of the polyp, but darker echogenicity in the lower end. It was not clear where the polyp arose from, as it was both attached to the submucosal layer of the esophagus and came from tissue deeper than the muscularis propria. Fine-needle aspiration for cytology showed no malignancy. The patient refused removal of the polyp because he had no symptoms such as dysphagia, vomiting, or regurgitation. At his annual follow-up with EGD, the polyp had showed no

progression. Six years later, he had symptoms of dysphagia and chronic cough. We repeated imaging on the patient. The EGD found polyp progression that protruded into the stomach (Fig. 2). Computed tomography of the chest showed a long intraluminal esophageal mass 2.4x4.6 cm in thickness and 20 cm in length, arising from mid to lower esophagus, showing an extreme increase in polyp size. We attempted to remove the polyp by EGD but failed due to the polyp protruding into the stomach and it being unable to be removed through the gastro-esophageal junction. We altered the plan to remove it by esophagotomy at the cervical esophagus. A linear incision was made along the left cervical esophagus and the esophagus was opened. The stalk of the polyp was located, the polyp excised, and the esophagus and incision closed (Fig. 3). Barium swallowing after surgery showed no contrast leakage from esophagus (Fig. 4). One week after surgery the patient was discharged without any complications. Pathology reported an atypical lipomatous tumor of the esophagus. Follow-up one year later showed no recurrence.

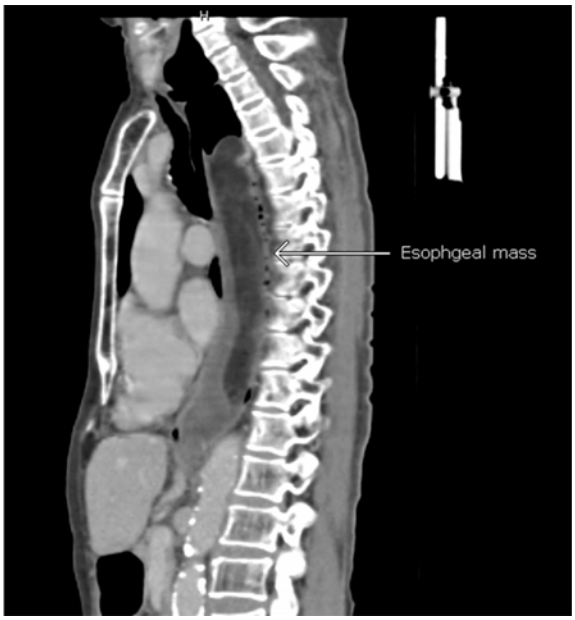

Fig. 1 CT scan show intraluminal mass in esophagus.

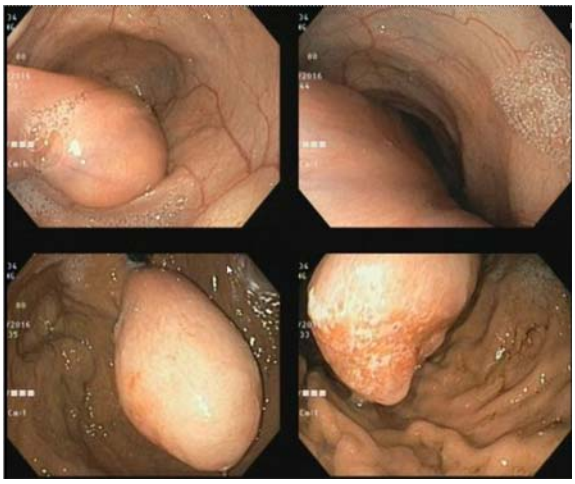

Fig. 2 EGD show polyp protrude into stomach.

Discussion

Several classifications of benign esophageal tumors are known, but the simple way to differentiate them is the origination of tissue layers of the esophagus. Although fibrovascular polyps are the most common intramural benign tumor of the esophagus, incidence is less than 1%[2,3] and they are difficult to differentiate from atypical lipomatous tumors because gross lesions are similar in appearance. Clinical presentation depends on the size of tumor, differences of asymptomatic patients, obstruction, compression of adjacent tissue, regurgitation, and bleeding. Fifty-four percent of patients presenting with dysphagia and regurgitation[2] are in danger of sudden mortality due to asphyxiation[4]. Removal of polyps as soon as possible has been recommended[5]. Due to esophageal peristalsis, polyps can progress along the esophageal lumen like a sausage lesion. The diagnosis evaluation includes contrast esophagography, computed tomographic (CT) scan, magnetic resonance imaging (MRI), endoscopy, and endoscopic ultrasound (EUS). Esophagography demonstrates sausage-like lesions, CT scans and MRI show components of adipose and fibrous tissue[6]. The most common mass is a large elongated polyp with a smooth surface, meaning that the tumor's length is approximately 14 cm and attached to the cervical esophagus just below the cricoesophagus[7]. For a typical lipomatous tumor and fibrovascular polyp, the gross lesion is similar but atypical lipomatous polyps are locally aggressive, with mesenchymal neoplasm composed with potential local recurrence and will need long-term follow-up. For correct diagnosis immunostaining such as MDM2, CDK4, or FISH gene amplification should be used for examination[7]. Many surgical techniques are used for tumor removal, including endoscopic resection[8-10]. In large tumors or when there is a lot of blood flow at the stalk, the standard open approach is necessary, with longitudinal esophagotomy followed by ligated

Fig. 3 Esophagotomy at cervical esophagus and polyp.

Fig. 4 Barium swallowing after surgery.

resection of the tumor stalk and 2-layer closure. Our patient was under surveillance for prostate cancer showing a lesion at the esophagus. At the time of presentation, he had no symptoms and refused removal of the tumor although he was at risk of sudden death. After annual follow-ups, he still had no symptoms until six years later. He had obstructive symptoms and the tumor could not be removed by endoscopy. We used open approach to remove the tumor, which was successful without complications. This case demonstrates that for patients with no symptoms, it is possible to delay surgery without complications and with a lower risk of asphyxiation. Standard open approach can be performed by general surgeons who do not have access to endoscopy equipment or are based in rural hospitals. This approach is simple and safe to remove the esophageal polyp.

What is already known on this topic?

Atypical lipomatous tumor is a large intraluminal polyp that presents itself similar to a large fibrovascular polyp, which is difficult to differentiate until pathological examination. For correct diagnosis immunostaining such as MDM2, CDK4, or FISH gene amplification should be used for examination. Removal of polyps as soon as possible has been recommended for prevent complication. Removal of the tumor can be performed by minimally-invasive surgery or open approach with free margin.

What this study adds?

This case demonstrates that for patients who have no symptoms, it is possible to delay surgery without complications and with a lower risk of asphyxiation. Standard open approach can be performed by general surgeons who do not have access to endoscopy equipment or are based in rural hospitals. This approach is simple and safe to remove the esophageal polyp.

Potential conflicts of interest

None.

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การผ่าตัด Giant pedunculated atypical lipomatous tumor ที่หลอดอาหารสามารถรอการผ่าตัดหลังจากวินิจฉัยได้ 6 ปี โดยไม่พบภาวะแทรกซ้อน

ปีติโชติ หิรัญเทพ, ไชยรัตน์ ทรัพย์สมุทรชัย, ณัฐพล อาภรณ์สุจริตกุล, จักรพันธ์ จิรสิริธรรม, ธีรวุฒิ รักชอบ, นามศิริ ใบอดุลย์

เนื้องอกที่ไม่ใช่มะเร็งของหลอดอาหารมักจะไม่ค่อยแสดงอาการจนกว่าจะมีขนาดใหญ่ที่พบได้บ่อยคือ fibrovascular polyp ซึ่งโดยลักษณะภายนอกจะแยกได้ยากจาก atypical lipomatous tumor ซึ่งเป็นชนิดที่มีการเป็นซ้ำสูงและอาการแสดงเหมือนกันต้องใช้วิธีการตรวจทางห้องปฏิบัติการถึงสามารถแยกออกจากกันได้ โดยทั่วไปถ้าตรวจพบมักแนะนำให้ทำการตัดเนื้องอกชนิดนี้ทิ้งทันทีเพราะมีโอกาสที่จะเสียชีวิตได้จากภาวะแทรกซ้อนที่จะอุดกั้นทางเดินหายใจ รายงานฉบับนี้พบผู้ป่วยที่เป็นเนื้องอกชนิดดังกล่าวขนาดใหญ่แต่ไม่ได้ทำการตัดในทันทีโดยที่ติดตามการรักษามาหกปี และได้มาทำการผ่าตัดเอาออกด้วยวิธีการตัดเปิดหลอดอาหารและเย็บปิดเนื่องจากขนาดที่ใหญ่ทำให้ไม่สามารถทำการเอาออกโดยวิธีการส่องกล้องผ่าตัดได้ ซึ่งวิธีการผ่าตัดแบบเปิดหลอดอาหารเป็นวิธีที่ง่ายและปลอดภัยสามารถใช้ในเป็นทางเลือกที่ศัลยแพทย์ควรทำในสถานการณ์ที่ไม่สามารถทำการผ่าตัดผ่านกล้องหรือไม่มีเครื่องมือที่เพียงพอได้
