Recurrent giant fibrovascular polyp of the esophagus

Ser Yee Lee, Weng Hoong Chan, Ranjiv Sivanandan, Dennis Teck Hock Lim, Wai Keong Wong

Abstract
Giant fibrovascular polyps of the esophagus and hypopharynx are rare benign esophageal tumors. They arise most commonly in the upper esophagus and may, rarely, originate in the hypopharynx. They can vary significantly in size. Even though they are benign, they may be lethal due to either bleeding or, rarely, asphyxiation if a large polyp is regurgitated. Patients commonly present with dysphagia or hematemesis. The polyps may not be well visualized on endoscopy and imaging plays a vital role in aiding diagnosis as well as providing important information for pre-operative planning, such as the location of the pedicle, the vascularity of the polyp and the tissue elements of the mass. They can also be recurrent in rare cases, especially if the resection margins of the base are involved. We review the recent literature and report a case of a 61-year-old man with a recurrent giant esophageal fibrovascular polyp with illustrative contrast barium swallow, CT and intra-operative images, who required several surgeries via a combination of endoscopic, trans-oral, trans-cervical, trans-thoracic and trans-abdominal approaches.

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Key words: Giant fibrovascular polyp; Esophagus; Esophageal polyp; Fibroepithelial polyps

Peer reviewer: Leonidas G Koniaris, Professor, Alan Livingstone Chair in Surgical Oncology, 3550 Sylvester Comprehensive Cancer Center (310T), 1475 NW 12th Ave., Miami, FL 33136, United States

INTRODUCTION
Giant fibrovascular polyps of the esophagus are rare. Even though they are benign, they may be lethal due to either bleeding or asphyxiation if regurgitated. Recurrence of these giant polyps are rare and treatment is often difficult, specialized and may require combination of a few treatment approaches and modalities including endoscopy and open surgical techniques. We illustrate this problem with a case report and review the literature.

CASE REPORT
A 61-year-old man with a medical history of hypertension and hyperlipidemia presented with melana and symptomatic anemia. His initial hemoglobin was 6.2 g/dL. He was transfused appropriately and underwent an emergent esophago-gastro-duodenoscopy (OGD). The endoscopy revealed a large polyp, arising from the upper esophagus settling in the upper stomach (Figure 1). The endoscopic biopsy revealed benign squamous mucosa and granulation tissue. He was offered surgical excision of the polyp in view of the bleeding episodes from the ulcerated polyp but he declined surgery. He was initially able to regreturate polyps out into the mouth, but he gradually progressed to mild intermittent dysphagia and agreed to surgical intervention 6 mo after initial presentation.

Under general anesthesia, laryngoscopic and esophagoscopy was performed. The polyps were noted at the cardio-esophageal junction but it was not possible to pull the polyps with their long stalks into the stomach. The large polyps were noted at the cardio-esophageal junction but it was not possible to pull the polyps with their long stalks into the stomach for adequate resection. In view of these difficulties, we proceeded with esophagotomy via a left thoracotomy approach to deliver and excise the polyps.

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Final histology revealed two fibrovascular polyps with ulceration measuring 4 cm × 4 cm and 6 cm × 6 cm, respectively. There was no evidence of malignancy. He was discharged in good health and followed up in the outpatient clinic at 3-6 mo intervals. He developed gradual intermittent dysphagia again 2 years later. A flexible OGD was performed and it revealed a recurrent fibrovascular polyp. In this admission, peroral excision of the recurrent polyp was performed with the aid of an ENDO GIA™ 30 stapler (Ethicon, Cincinnati, OH). The esophageal lumen was then inspected endoscopically and found to be free from bleeding, mucosal tears, or perforation prior the end of the procedure.

Histology was consistent with a previous report of a giant fibrovascular polyp, measuring 5 cm × 6 cm. There was no evidence of malignancy, however, the resection margin was noted to be involved. He was discharged in good health and followed up in the outpatient clinic at 3-6 mo intervals.

He re-presented approximately 2 years later, again for similar symptoms of intermittent dysphagia. An examination under anesthesia with a direct laryngoscopy and a rigid esophagoscopy revealed another polyp with a long stalk arising from the upper esophagus. In view of the recurrent nature and the previous surgeries, a barium swallow and CT of the neck and thorax were performed to aid surgical planning (Figure 2).

In this second recurrent episode, intra-operative findings revealed a 6 cm × 7 cm polyp with a long stalk measuring about 5 cm and a broad base of 1 cm width originating from the upper third of the esophagus. A trans-cervical esophagotomy was performed initially but due to the size of the polyp, it was not possible to deliver the polyp and an upper esophagotomy via a right thoracostomy approach was performed to deliver and resect the polyp. Excision of the polyp via the thoracic esophagotomy and excision of fibro-vascular stalk via the cervical esophagotomy was performed (Figure 2). Histology was consistent with that of a giant fibrovascular polyp with no evidence of malignancy. He was discharged and is well to date.

**DISCUSSION**

Fibrovascular polyps of the esophagus are rare benign tumors, comprising about 1% of all benign esophageal tumors, however, they are the most common intraluminal benign tumors of the esophagus[1]. Giant fibrovascular polyps are defined as polyps larger than 5 cm in maximum diameter. To date, there are just over 100 reported cases in the literature and the largest single series consists of 16 patients[2,3]. They are slow growing, pedunculated tumor masses that often arise from the upper esophagus, near the level of the cricopharyngeus at the pharyngo-esophageal junction, and this area has also been termed as the Laimer-Haeckermann triangle (also known as the Laimer's triangle)[4]. The pathogenesis of these polyps is thought to originate from the loose and redundant submucosal tissue near the Laimer's triangle. This relatively mobile tissue due to lack of muscular support, through years of esophageal peristalsis traction and swallowing, is dragged along, elongated and enlarged intraluminally.

The histology of the fibrovascular polyp consists of a mixture of lipomatous tissue among dense or loose fibrous elements, accompanied by an abundant network of vessels and covered by a normal squamous epithelium. The squamous epithelium may ulcerate and bleed especially in the larger tumors. Particularly in giant polyps, different histological components may vary and one may predominate leading to different terminology of these lesions as fibroepithelial polyps, fibrolipomas, fibromyxomas, lipomas, fibromas. These are all now collectively classified by World Health Organisation as
fibrovascular polyps. The more common differential diagnosis includes leiomyomas, leiomyosarcomas, squamous papillomas, lymphomas, spindle cell carcinomas and hemangiomas amongst others.

Malignant transformation is rare but has been reported in esophageal polyps. The lipomatous components can undergo sarcomatous changes, the squamous mucosa can develop into squamous carcinomas and small polyps have developed into adenocarcinoma[1].

Due to the indolent nature of these polyps and the potential space the esophagus provides, these fibrovascular polyps can grow up to considerable sizes without causing many symptoms till late, measuring as big as 26 cm in largest diameter[2]. The majority of fibrovascular polyps occur in elderly men aged between 60 and 70 years old, but they has been reported in a 5-mo-old infant[3].

Though biologically benign, these giant fibrovascular polyps can have dramatic and even life-threatening presentations. Due to their size and mobility, they can be regurgitated and can cause asphyxiation or require emergent airway management[4]. In the literature, dysphagia was the most common complaint (present in 87%), followed by respiratory symptoms (25%) and regurgitation of the polyp into the pharynx or mouth (12%). The other reported non-specific symptoms included epigastric pain, odynophagia, non-exertional substernal chest pain, loss of weight, persistent cough and in our case, gastrointestinal bleeding[5,6].

In the absence of an obvious regurgitation of the polyp during examination, diagnosis can be a challenge[7]. During endoscopy, the lesion may be missed especially if the origin of the stalk is not visualized, as the polyp may occupy the esophageal lumen and the surface of the polyp can resemble normal esophageal mucosa[8]. Diagnosis can be made by a combination of clinical history and various investigations such as endoscopy and imaging studies like barium swallow studies, endoscopic ultrasonography (EUS), CT and magnetic resonance imaging (MRI). Barium studies are commonly used and can show up the characteristic appearance of a smooth intra-luminal sausage-shaped mass with bulbous tips, with varying degrees of lobulation (Figure 1)[9]. It must be noted that, on occasion, these polyps may be opposed against the esophageal wall and give a false impression of a normal barium swallow. EUS can be a useful adjunct as it provides information on the size, origin of the stalk and vascularity of the polyp. In addition, EUS-fine needle aspiration may provide a more diagnostic histological sample than a superficial biopsy from an endoscopic approach. CT has been recommended by some; in addition to providing information on the components of the tumor, at an early arterial phase, especially in large polyps, feeding vessels can be visualized which can aid in surgical planning. In such cases, where a large feeding vessel is demonstrated, open surgery is advocated instead of endoscopic techniques e.g. snare polypectomy[10]. MRI is also a useful adjunct in diagnosis and surgical planning as it provides multiple planes of section and high soft tissue differentiation resolution[11].

The mainstay of treatment once diagnosis is achieved is surgical excision in view of the potential risk of respiratory compromise, bleeding (as in this patient) and the debilitating symptoms. Surgery also serves to exclude cancer and avoid the small risk of malignant degeneration. Depending on the size, the location of the stalk's base and its mobility, different approaches have been practiced. The different methods include simple endoscopic excision techniques using electrocautery or even Nd:YAG laser ablation in a case, cervical esophagotomy, trans-thoracic esophagotomy and esophagectomy[12]. Smaller polyps, less than 2 cm in diameter with a thin pedicle, can be removed endoscopically without many complications but this is not recommended in larger tumors (length > 8 cm). Due to the thick vascularized pedicle, hemostasis is most safely achieved by open surgical techniques[13]. As the origin of the pedicle of the polyp in the majority of the cases lies in the upper third of the esophagus, cervical esophagotomy seems like the approach of choice. However, it is important take note, at this point, the value and accuracy of pre-operative assessment of the base of the pedicle and the bulk of the tumor mass. Occasionally, a thoracotomy should be considered as it may be required for difficult, large lesions or lesions with a low origin. On occasion, a trans-abdominal approach via laparotomy may be a useful option as well, to aid in the delivery of the bulky polyp head through a gastrostomy[14].

We advocate an individualized surgical strategy according to the characteristics of each polyp and recommend that in difficult cases [recurrent cases, patients with more than one polyp or patients with a large polyp (> 5 cm) with a long stalk (> 5 cm)] esophagotomy via thoracotomy should be considered for good control of hemostasis as well as for providing adequate exposure for resection of the pedicle's origin and any redundant mucosa around the pedicle. Recurrence of giant fibrovascular polyps is rare but has been described[12,13]. We believe that residual tissue around the pedicle's base may cause recurrent polyp formation which we hypothesize is the reason for the recurrent nature of polyp formation in our patient.

We therefore suggest appropriate pre-operative counseling and investigations such as obtaining additional informed consent for thoracotomy and laparotomy procedures. We also recommend patients who smoke or have significant respiratory morbidity should have a pre-operative lung function assessment. Peri-operatively it is also essential to inform the anesthetist and operating theatre staff and prepare the patient in anticipation for a thoracotomy or a laparotomy should the situation arise.

In summary, giant fibrovascular polyps are rare esophageal tumors and recurrences are even more uncommon. Although most are benign, surgical excision is recommended in view of potential deadly complications e.g. asphyxiation, bleeding, malignancy. Adequate pre-operative investigations should aim to identify the pedicle's origin and the bulk of the polyp to aid the planning of the surgical approach.
to ensure adequate hemostasis and clear resection of the base to prevent recurrences. The surgical options include endoscopic resection, open surgery via trans-cervical, trans-thoracic or trans-abdominal approaches. This should be tailored on a case-to-case basis and a combination of the approaches should be considered in difficult cases.

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