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Unusual presentation of a giant benign inflammatory polyp in the upper esophagus



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ABSTRACT

INTRODUCTION: Benign inflammatory fibroid polyps (IFP) are rare submucosal tumors of the upper gastrointestinal tract. Rarely, they can develop in the esophagus, usually in the lower third. There are only 12 cases of giant IFP of the esophagus reported in literature and little is known about their origin, biological behavior and operative management. We present a patient with a giant benign IFP of the esophagus that originated from the upper esophagus.

CASE PRESENTATION: The patient is a 59-year-old male who presented with dysphagia. Upper endoscopy and esophagram revealed a giant intraluminal esophageal mass with a pedicle in the upper esophagus. Resection of this mass was performed through a left cervical esophagotomy. Pathology confirmed IFP, On 2 year follow up, there was no recurrence of the mass.

DISCUSSION: A giant IFP is defined as an IFP greater than 4 cm, commonly present in the distal esophagus. Pathology usually reveals vascularized fibrous stroma with elements of inflammatory infiltrate. This mass is slow-growing and asymptomatic until it grows to a large size. Common diagnostic studies include barium esophagram, upper endoscopy, and CT imaging. A key pre-operative work-up is to identify the location of the pedicle to plan out surgical approach and to avoid injuring the rich blood supply thus preventing a life threatening hemorrhage during the operation.

CONCLUSION: Giant IFPs are infrequent in clinical practice. Resection is indicated and usually performed by a surgical intervention or endoscopic removal. The pathogenesis of these polyps remains poorly understood due to the rarity of these lesions.

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1. Introduction

Inflammatory fibroid polyps (IFP) of the esophagus are very rare. Giant IFPs are even more rare. Only 12 case reports of giant IFP has been published in literature. Most of these polyps have been reported in patients in their 5th and 6th decades of life. Owing to the rarity of these polyps, little is known about their origin, biological behavior and operative management.

The majority of these benign esophageal polyps are located in the distal third of the esophagus [1]. They frequently occur as solitary lesions and may vary greatly in size [2]. These masses are slow growing and present for many years as an asymptomatic mass. As the mass enlarges, it can result in symptoms such as heartburn, regurgitation, and most commonly dysphagia [1]. Barium studies and upper endoscopy can be used for the diagnosis, but the definitive diagnosis for IFP is histological. Depending on the size of the polyp, endoscopic or surgical resection is recommended.

In this paper, we present a rare case of a 12 cm giant IFP that originated from the cervical esophagus. To the best of our knowledge, this is only the second reported case of an IFP originating from the cervical esophagus. This patient underwent polyp resection via a left cervical esophagotomy.

2. Case description

A 58-year-old male presented with progressive dysphagia for 2 years. The patient denied chest pain or weight loss. Barium esophagram revealed an obstructive filling defect in the proximal esophagus (Fig. 1). Upper endoscopy and endoscopic ultrasound showed a large pedunculated polypoid mass that was obstructing the esophageal lumen from 20 cm to 40 cm from the incisors, with the distal end of the tumor intermittently prolapsing through the gastroesophageal (GE) junction into the stomach on retroflex view. On endoscopic ultrasonography, the mass was involving the submucosal layers. The stalk of the polyp was originating from the cervical esophagus, just below the cricopharyngeous muscle. A chest Computed Tomography (CT) scan demonstrated a large lobulated mass occupying nearly the entire length of the thoracic

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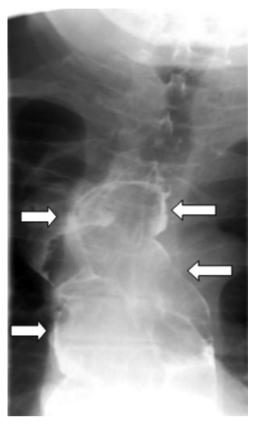


Fig. 1. Barium esophagram demonstrating large filling defect in esophagus (arrow pointing to filling defect).

esophagus with a focal mass effect on the left atrium and trachea (Fig. 2). There was no lymphadenopathy.

The patient underwent a left cervical exploration and cervical esophagotomy (Fig. 3). The large polyp was delivered through the esophagotomy, and the pedicle was ligated and subsequently excised. The esophagus was repaired with a two layer closure and the left sternocleidomastoid muscle was used to buttress the repair.

Macroscopically, the specimen measured $12.0 \times 4.0 \times 3.0\,\mathrm{cm}$ (Fig. 4). The apical surface of the polypoid structure shows a focal area of ulceration and adherent necrotic tissue measuring up to 1.5 cm in maximal dimension.

Histological examination revealed vascularized fibrous stroma with patchy chronic inflammatory infiltrate with an overlying benign stratified squamous esophageal mucosa loose fibro-myxoid stroma with cytologically bland spindle cells, and scattered inflam-

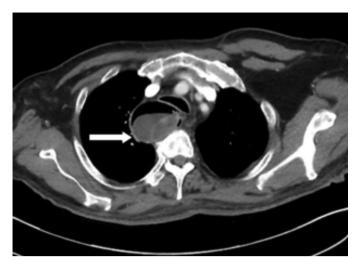


Fig. 2. Axial CT imaging demonstrating giant esophageal mass obstructing lumen and compressing trachea.

matory cells (Fig. 5). There were no signs of dysplasia or malignancy. These features were consistent with the diagnosis of IFP. On 2 year follow-up after resection, the patient had routine upper endoscopy which revealed no recurrence of the mass.

3. Discussion

Giant esophageal polyps are a rare entity with very few cases reported in literature. Depending on the histologic predominance, these lesions have been variously called fibroma, fibrolipoma, myxofibroma. True pedunculated esophageal polyps are mainly divided into two types, fibrovascular polyps (FVP) and less commonly IFP [3]. Most of the reported cases are FVPs that typically occur in the upper esophagus while inflammatory polyps occur in the distal esophagus. Currently, there are only 12 cases of Giant IFPs of the esophagus reported in the literature and little is known about their origin, biological behavior and operative management.

IFPs are typically small, submucosal, non-capsulated, pedunculated lesions; they can be found in small bowel and rarely in colon but found most commonly in the stomach [4]. IFPs occur very infrequently in the esophageal wall and when they do occur, these lesions are mostly found in the lower esophagus [5,6]. Rarely, IFP grows larger than 4cm and is defined as a giant IFP. Acid reflux injury is thought to play a role in the development of these lesions [7]. However, the exact pathogenesis of the IP is unknown. It is generally accepted that this is not a neoplasm, but is a reactive process to physical, chemical or microbiological stimuli [3].

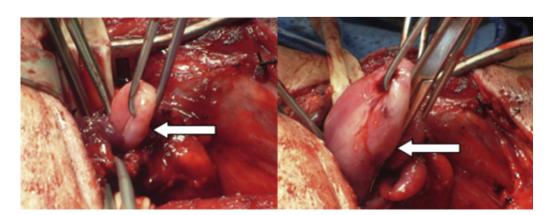


Fig. 3. Intraoperative photograph showing extraction of giant inflammatory polyp from the esophagus through a left cervical incision.

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Fig. 4. Gross specimen of giant inflammatory polyp, largest dimension 12 cm.

These patients typically present with dysphagia, which is the most common symptom [1]. The dysphagia is usually progressive, starting with solids and then advancing to liquids as the mass increases in size [1]. The regurgitation of the mass and food, and consequent asphyxiation has been rarely reported as well.

The diagnosis of IFP can be challenging and can sometimes be missed on upper endoscopy if small. Barium studies were found to be the most common initial diagnostic tool to diagnose esophageal polyps [8]. These studies reveal an intraluminal contrast filling defect within a widened esophagus. Endoscopy can provide valuable information especially in combination with endoscopic ultrasound, providing depth of esophageal involvement and the location of the stalk. Computed tomography and magnetic resonance imaging can give added information in evaluating these lesions. The diagnosis is secured with a histological specimen.

IFP excision can be performed with either an endoscopic approach or open surgical resection depending on size, location, and expertise of the surgeon or gastroenterologist. Occasionally, endoscopic resection of a giant IFP has been reported in the literature [9]. Endoscopic resection is reserved for the experienced clinician. The decision for endoscopic resection is based on the clinician's ability to completely excise the polyp and avoid catastrophic bleeding. Polyps with large broad bases are difficult to remove completely using endoscopic loops and therefore would not be amenable to complete endoscopic resection. Furthermore,

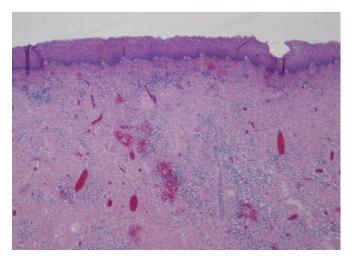


Fig. 5. Histological features of giant inflammatory polyp demonstrating mixture of large vascular structures, fibrous connective and myxoid tissue.

with the endoscopic approach, control of bleeding can be difficult and can lead to catastrophic hemorrhage. The EUS can provide important information pre-operatively about the vascularity of the polyp; the patients with large or complex feeding vessels should be directed toward surgical consultation. Equipment for hemostasis, including hemoclips, Coagrasper, injection needle and adrenaline for injection should be prepared before the procedure in case one encounters brisk bleeding. Another potential pitfall with endoscopic removal of polyps is the risk of burning and perforating the esophagus. This risk can be minimized with adequate insufflation and distension of the esophagus. The site of snare polypectomy should be kept away from the base of stalk to prevent esophageal perforation [10]. Our patient underwent a left cervical exploration with resection of the mass due to its large size. Knowing the location of the pedicle of the esophageal polyp is a crucial step in planning for resection. This knowledge can be provided, preoperatively, by performing a thorough and careful esophagoscopy. The location of the stalk dictates the surgical approach, either via transcervical, right thoracotomy, left thoracotomy or laparotomy. Since the pedicle has to be resected under direct vision, the incision and the esophagotomy has to be made opposite to the base of the polyp stalk [11]. Performing the esophagotomy on the side where the polyp originates can potentially cause severe hemorrhage [11].

4. Conclusion

Giant inflammatory fibroid polyps of the esophagus are extremely rare and usually occur in the distal esophagus. This case demonstrates that these polyps can exist in the upper esophagus and can grow to an extremely large size, causing severe symptoms. Resection is indicated in all patients to (1) confirm the diagnosis and (2) to reduce symptoms as well as prevent a potential fatal asphyxiation event.

Conflicts of Interest

None.

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Consent

Consent form has been filled and signed by the patient.

Author contribution

Badi Rawashdeh: study design, writing.

Mark Meyer: study design.

Mohammad Moslemi: data collections.

Jasmine Gill: data collections.

Samuel Kim: writing.

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