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Case Report

Giant Inflammatory Fibroid Polyp of Upper Esophagus: A Life - Threatening Rare Case

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1. Abstract

Inflammatory fibroid polyps IFPs of the esophagus are very uncommon. Giant IFPs are an extremely rare entity and the majority of them are located in the distal third of the esophagus. They are slowly growing and remain clinically "silent". As the mass enlarges clinical symptoms appear with dysphagia, chest pain and food regurgitation being the most common. Their clinical course might be indolent and acute respiratory distress might be a life-threatening clinical manifestation of them. We present a case of a 54 years old woman suffering from dysphagia for at least 6 months. Upper gastrointestinal endoscopy revealed a giant esophageal polyp extending from the cervical esophagus almost to the thoracic esophagus. A hot snare polypectomy was attempted, but the polyp prolapsed through the mouth and caused acute respiratory distress. The patient was transferred to the Otolaryngology (OR) department, where urgent orotracheal intubation was performed, using a CMAC video laryngoscope, and the tumor was successfully removed via a transoral approach. Giant IFPs are infrequent in clinical practice and they usually occur in the distal esophagus. The pathogenesis of these polyps remains poorly understood. Because of the risk of lethal complication (asphyxia), it is strongly recommended to remove them once they are diagnosed. These tumors can be removed through transoral, transcervical, transthoracic and endoscopic approaches depending on the location and the size of the tumor. The impressive size of gigantic polyps makes their treatment challenging for surgeons and gastroenterologists.

2. Introduction

Inflammatory Fibroid Polyps (IFPs) are rare benign tumors of the gastrointestinal tract. Giant IFPs are even more uncommon in clinical practice [1]. They most commonly appear in the stomach. Only a few reports of giant IFPs have been published [1] and the majority of these polyps occur in the distal esophagus. Their clinical course might be indolent [2]. As they grow in size, they can provoke intense symptoms such as dysphagia, heartburn and food regurgitation, or even gastrointestinal bleeding and respiratory distress [1, 2]. Upper gastrointestinal endoscopy remains the gold standard for the diagnosis although the histological examination will confirm the type of the lesion [3, 4]. According to the size and location of the polyp, endoscopic or surgical approach can be performed to remove it. In this paper, we present a life-threatening case of a giant IFP originating from the cervical esophagus causing acute respiratory distress.

3. Case Report

A 54 years old woman presented to the Gastroenterology department for examination. She was suffering from progressive dysphagia in liquid and solid food as well as often regurgitations for at least 6 months. She had a medical history of cholecystectomy and nephrolithiasis. She was also a heavy smoker (40 pack years) and obese (BMI: 34).

During the last 6 months, she was examined by a psychiatrist, a general practitioner and a neurologist due to often vomiting without weight loss and symptoms of depression and was prescribed antidepressants with no improvement of the symptoms.

She underwent an upper GI endoscopy which revealed a large (giant) pedunculated polyp extending from the cervical esophagus to the lower esophageal sphincter. An endoscopic hot snare polypectomy was decided. Initial attempt of endoscopic resection after clipping the stalk very near to the base led to regurgitation of the mass and intraoral prolapse of the polyp, causing respiratory distress and cyanosis. Attempts to push back the mass through the esophagus failed and finally because of the imminent airway obstruction, a large Kocher surgical forceps was used to catch the giant polyp in tension outside the mouth and preserve a portion of upper respiratory tract, in order to be avoided the asphyxia of the patient (Figure 1). The patient was transferred to the ENT surgery room, where urgent orotracheal intubation, using a CMAC video laryngoscope, was performed. The woman underwent a direct hypopharyngoscopy. Sutures were placed in the polyp's base and the mass was removed with a transoral approach using metal surgical instruments with no need for electrocautery of the polyp's base (Figure 2). The postoperative period was uneventful and the woman was discharged on the 7th postoperative day. The larger resected specimen measured 8.6x3.1x2.1 cm and histological examination showed vascularized fibrous stroma with hyperplastic changes of the epithelium and the connective tissue, with infiltration of inflammatory cells such as lymphocytes, eoshinophils and plasma cells leading to a diagnosis of Giant Inflammatory fibroid polyp (Figure 3). A follow up endoscopy was performed 6 months and one year later, with no recurrence of the polyp (Figure 4).

Figure 1: Polyp mass grasped and stretched out of the mouth in order to avoid airway obstruction.

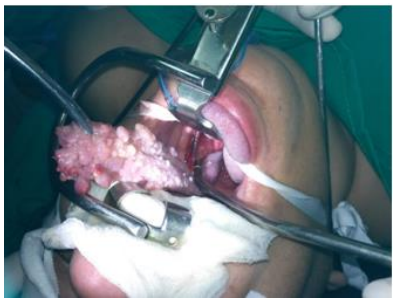

Figure 2: Removing the polyp mass with a transoral approach under general anesthesia.

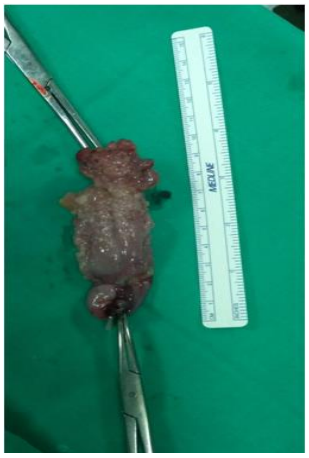

Figure 3: Surgical specimen measuring 8.6 cm sent for histopathologic examination.

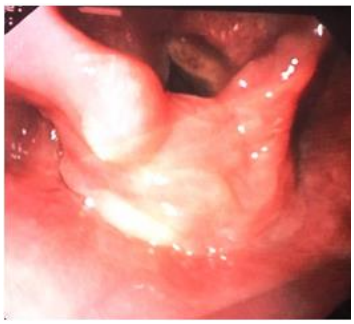

Figure 4: Follow up endoscopy after 6 months with no findings of recurrence of the polyp.

4. Discussion

In 1949 Vanek et al first described a "gastric submucosal granuloma with eosinophilic infiltration" [5]. Four years later Helwig and Ranier used the term Inflammatory Fibroid Polyps (IFPs) to describe them [6]. IFPs are usually small in size, submucosal, pedunculated, benign lesions most commonly located in the stomach and rarely in the colon and the small bowel and more rarely in the esophagus [7-9]. IFPs can become larger than 4 cm and when they do, they are called giant IFP [10, 11]. On histology IFP is an inflammatory mass characterized by involvement of the submucosa and lamina propria, hyperplasia of connective tissue, hyperplasia of small-sized vessels, concentric arrangement of fibrous connective tissue and inflitration with inflammatory cells (eosinophils, plasma cells, lymphocytes).

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Many etiologies have been suggested, but their pathogenesis is still unknown, although the inflammatory theory is the most dominant. Some authors suggest an allergic reaction due to the abudance of eoshinophils on histology, while others implicate factors such as neural hyperplasia, trauma, bacterial or chemical stimulants [12]. Acid reflux might also be responsible for their development [13].

In the literature, there are no more than 15 described cases of giant IFPs of esophagus and most of them are located in the lower or middle esophagus. IFPs of esophagus might have an indolent clinical course and be asymptomatic for years, as they are slowly growing [2]. The clinical presentation depends on their location and size. They might present with symptoms such as dysphagia, heartburn, regurgitation and more rarely gastrointestinal bleeding or even respiratory distress [1, 14]. To our knowledge this is the third case of a giant IFP of upper esophagus, and the first one to cause respiratory distress.

The diagnosis of IFP is challenging and most commonly is made post-surgically based on histology studies [1]. Upper GI endoscopy provides valuable information concerning the lesion and its characteristics. CT, MRI and Endoscopic Ultrasound (EUS) offer added information concerning the location, the size and the stalk of the mass.

The differential diagnosis of IFPs generally includes benign tumors such as schwannoma, GIST, leiomyoma, perineurioma, vascular neoplasms, melanoma, and inflammatory myofibroblastic tumor [10]. In our case, the main differential diagnosis due to the site of the mass appearance (upper esophagus) was giant fibro vascular polyp of esophagus (FVP) [15]. Unlike IFPs of esophagus which usually appear in lower esophagus, FVPs are masses that usually appear in upper esophagus and might cause respiratory distress as a presenting symptom. There are almost 120 cases in the literature of FVPs of esophagus. Differential diagnosis can be made with the help of histology specimens.

The treatment of IFPs is excision that can be performed by endoscopy, surgical with the help of endoscopy or open surgical method. These tumors can be removed through transoral, transcervical, transthoracic and endoscopic approaches based on the location and the size of the tumor. Small pedunculated polyps can be successfully removed with an endoscopic approach [16], while bigger ones or ones with large broad bases are difficult to be removed with endoscopic resection. In such cases, the risk of massive bleeding must be taken into consideration. It's up to the clinician's ability to finally decide whether to perform an endoscopic polypectomy or to follow an open approach, weighing the benefits and the risks of each method. In our case, an endoscopic approach was first attempted but because of the location of the polyp (upper esophagus) and the fact that it prolapsed through the mouth, a transoral surgical approach was finally performed. Since the polyp could be removed with direct hypopharyngoscopy, there was no need for cervicotomy or pharyngotomy.

5. Conclusion

Giant inflammatory polyps of the esophagus are an extremely rare entity and they usually occur in the distal esophagus [1, 14]. To our knowledge, this is the third reported case of an IFP originating from the upper esophagus and the first one presenting with respiratory distress. It is also the first one encountered with a combined approach of endoscopy and finally transoral excision after direct hypopharyngoscopy. IFPs might have a silent clinical course and become large enough before they cause symptoms. Although benign, giant IFPs of esophagus might be life-threatening causing respiratory distress, and when diagnosed their removal is highly recommended.

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