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Case report

Recurrent giant fibrovascualr oesophageal polyp: Benefits and pitfalls of a multimodal approach☆

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ABSTRACT

Fibrovascular polyp of the cervical esophagus represents about 0.5% to 1% of all benign oesophageal tumours. Usually asymptomatic, when FP protrudes into the oesophageal lumen, this may cause respiratory obstruction and provoke dysphagia, vomiting, dyspnoea, and retrosternal pain. In this article, we describe a multimodal approach in the treatment of a complex recurrent FP, for which surgical resection represents the safer and less invasive procedure.

1. Introduction

Oesophageal fibrovascular polyps (EFP) are rare, benign, intraluminal tumours located in the upper oesophageal tract. The most frequent clinical manifestation is dysphagia, followed by regurgitation, chest and intestinal bleeding, or the sensation of a mass in the throat [1]. Because polyps are mainly asymptomatic they can reach a considerable size and diagnosis is usually incidental. Resection is mandatory as sudden death for asphyxia has been reported [2]. Diagnostic and therapeutic management can be challenging and often requires multimodality radiologic, endoscopic and surgical strategies. The endoscopic approach has clear benefits in a standard, less complicated case, but in complex cases, open surgery may present as the only treatment option. We report a very rare case of recurrent triple giant pedunculated fibrovascular polyp composed of three bases of implantation in the oropharynx in which endoscopic treatment failed and then was successfully removed through lateral cervicotomy. The experience reported emphasizes the difficulties encountered in the endoscopic approach of the recurrent poli-pedunculated polyp, a challenging procedure with high haemorrhage risk when applied to giant polyps as reported in literature, and then finally removed with a surgical excision. Cervicotomy resection, although considered a safe procedure, is not without risks and complications. As we described, in our case a cervical fistula occurred which was treated in a conservative way.

2. Case report

We present a case of a 79-year-old man with a recurrent giant fibrovascular polyp presenting dysphagia and mass regurgitation through the mouth. The patient had a past medical history of an endoscopic resection of a EFP of 5 cm performed five years ago, after which the patient was lost to follow up. Patient's comorbidities reported were allergic asthma, hiatal hernia, hypercholesterolemia, hyperglycaemia and hyperuricemia in treatment with budesonide/formoterol, terbutaline, simvastatin and omeprazole. Physical examination was normal, and no signs of lymphadenopathy were found. Results from laboratory analysis were unremarkable, except for fibrinogen 814 mg/dl (range 150–450 mg/dl), glycaemia 129 mg/dl (range 70–105 mg/dl), total cholesterol 270 mg/dl (range 100–200 mg/dl), alkaline phosphatase 65 U/l (range 40–130 U/l), proteinemia 6,2 g/dl (range 6,3–8,7 g/dl). A CT scan with 3D reconstruction (Video 1) and an MRI of the neck and thorax (Fig. 1) showed an intraluminal oesophageal lesion originating from the left aryepiglottic fold and the ipsilateral pyriform sinus, with a large exophytic component extending to the middle esophagus with dimensions of approximately 60 × 55 × 135 mm. The lesion presented as a vascularized stump originating from a branch of the left superior laryngeal artery. The FDG PET/CT showed no metabolic activity. The endoscopic study confirmed the presence of an oesophageal polyp arising from the posterior arytenoid fossa and extending up to approximately 38 cm from the dental arch. A preoperative endoscopic biopsy

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revealed mesenchymal proliferation consisting of mature adipose tissue with fibrotic tracts and occasional multinucleated atypical cells (Fig. 2). A first endoscopic resection was planned but failed, as extracting the polyp through the mouth was not possible. Therefore, on a second endoscopic approach, an endoscopic loop was positioned around the pedicle of the polyp with the aim to provoke the ischemia of the lesion. However, the endoscopic double-loop strangulation of the polyp was unsuccessful. Consequently, a week later, an endoscopic-assisted left-lateral cervicotomy was performed allowing the isolation and resection of a triple oesophagic polyp (Fig. 3). The pedicle bases were resected using radiofrequency sealing (Fig. 4) and sutured with absorbable monofilament. The procedure was performed by E.V. and Y.Q, two specialized high experienced general surgeons. After surgery, the patient was admitted to the intensive care unit from which he was discharged on the fourth post-operative day to return to the surgical department. Here, results showed hyperpyrexia (37.7 °C) and leucocytosis (12,700/mm^3). Thus, on the fifth post-operative day, a CT scan revealed the presence of a hypodense collection approximately 15 cm wide in the anterior mediastinal and left paraesophageal site that extended towards the lower left cervical region, forming a dense lesion measuring 32 × 33 cm containing blebs, suggesting cervical fistula. The patient was treated with nil-by-mouth for seven days and with parenteral nutrition, and the collection was drained through the surgical wound. Furthermore, fasting was maintained with adequate antibiotic therapy. The patient was discharged on the twenty-fifth postoperative day after a resumption of normal eating. A control endoscopy confirmed the absence of the fistula, and the laboratory analysis returned normal values. We probably reconduct the forming of fistula to a suture leakage on one of the implantation bases of the polyp due to the patient's comorbidity (hypercholesterolemia, hyperglycaemia and hyperuricemia) and patient's corticosteroid treatment.

3. Discussion

EFP is a rare tumour of the pharyngoesophageal junction whose pathogenesis is a matter of discussion. Some authors claim that a lack of muscular support in the Laimer triangle may cause a progressive elongation of tissue due to peristalsis traction and swallowing [1]. On the contrary, other authors [3,4] who performed a cytogenetic study confirmed that EFP presents multiple complex chromosomal changes with signs of ring instability that could suggest that EFP is a neoplastic process rather than a consequence of redundant hamartomatous oesophageal tissue.

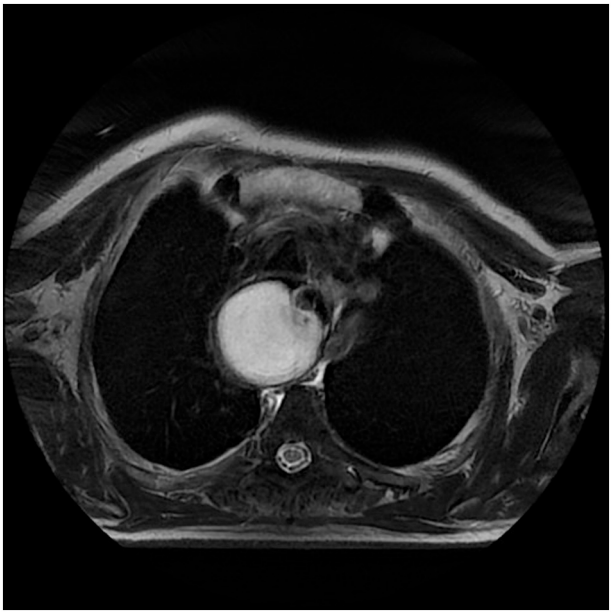

Fig. 1. MRI image.

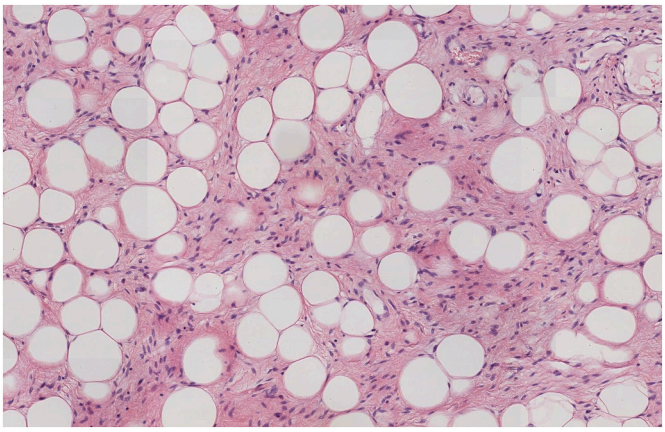

Fig. 2. Preoperative biopsy.

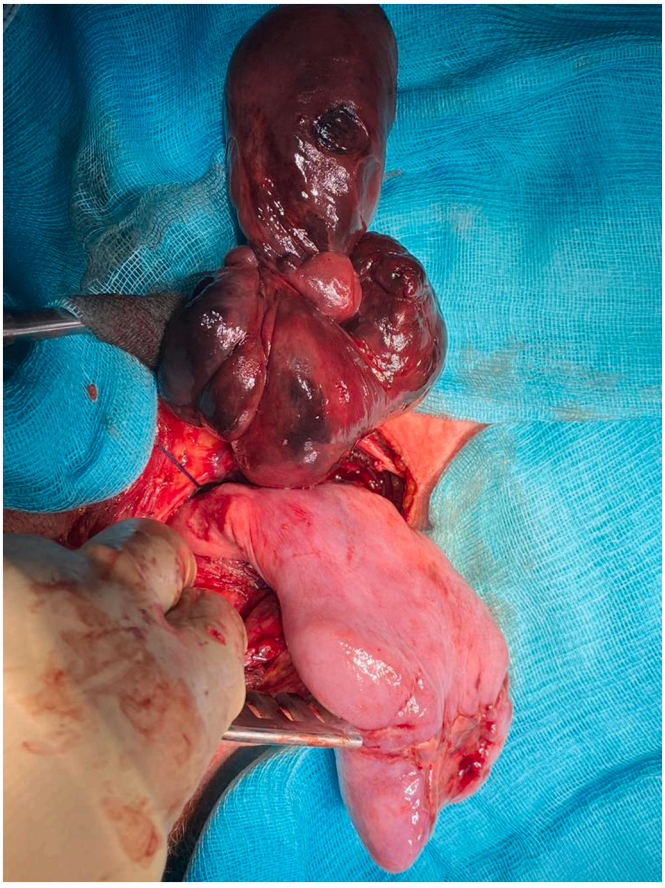

Fig. 3. Intraoperative field.

In this report, we described a complex case of a triple recurrent giant oesophageal polyp for which endoscopic resection and double-loop strangulation failed; thus, surgical approach became the only possible treatment option. The patient had previously submitted to an endoscopic resection five years ago, and was subsequently lost to follow up. In the literature, recurrent lesions are not common, although some cases have been reported [4]; however, in the majority of reported cases follow up has not been reported, which can lead the under-estimation of the recurrence rate.

The removal of giant pediculated fibrovascular polyps is usually

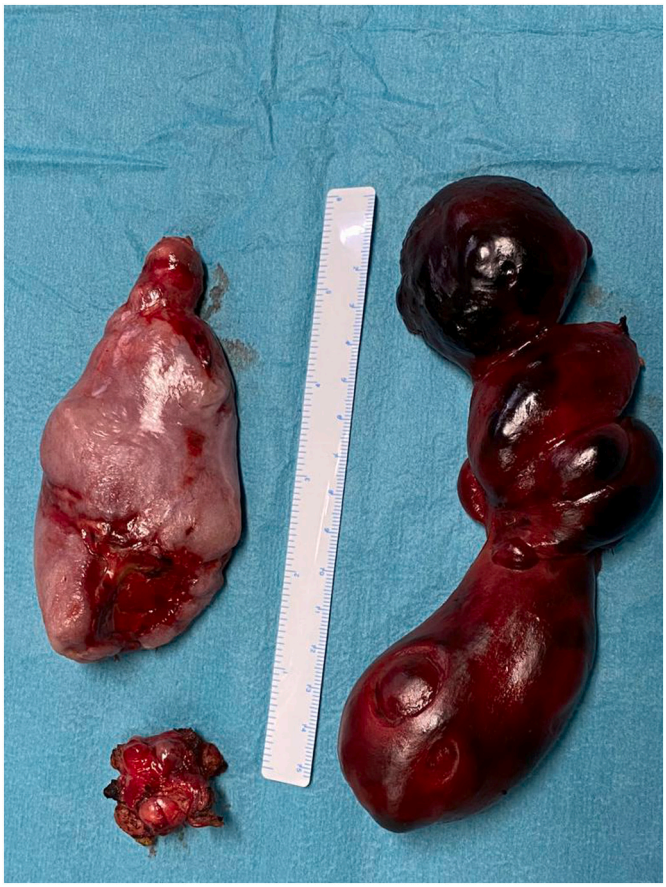

Fig. 4. Triple polyp resection.

accomplished by surgical or transluminal approach. Transluminal resection can be achieved by means of pure transoral access or by endoscopic support, and has been gaining wider acceptance over the last decade. Indeed, endoscopic resection represents a more conservative and less invasive approach, which may lead to a reduced length of hospital stay and better post-operative outcomes in terms of pain and possible complications. Therefore, this should be the first choice in the treatment of patients with EFP.

The endoscopic removal of EFP is challenging because of the difficulty of controlling the polyp stalk, though several studies have shown that this is also an effective approach in large polyps [5,6]. Nevertheless, in selected cases, such as the one presented here, surgical treatment remains the only possible option and oesophagotomy and polyp resection performed by a lateral cervicotomy is the most commonly used approach. Polyp resections via the trans-thoracic approach have been also described [7] and used for a large polyp with a high risk of airway compression. Moreover, a combined approach (cervicotomy/laparotomy/thoracotomy) has also been described due to the impossibility of retracting the polyp cranially, through either cervicotomy or thoracotomy [8]. In the literature, total esophagectomy has been reported in few cases [9] for the pre-operative misdiagnosis of an intraluminal tumour. With a larger polyp, if extraction through cervicotomy is not feasible, gastrotomy can be performed, as described in five patients, two of whom with the laparoscopic approach.

Patient selection is mandatory and accurate pre-operative studies, as in the case here, play a fundamental role: 3D reconstruction was performed and permits planification.

It is our view that endoscopic resection should be performed in all patients, due to the zero number of cases of stalk bleeding and esophagus perforation reported in the literature. Endoscopic removal has also been successful in the treatment of large polyps.

By a literature review, very few cases are reported of a recurrent giant polyp treated with cervicotomy.

Considering the resection methods like endoscopic ligation, esophagotmy and thoracotomy or the combined approach, the treatment of choice has to be related to polyp's carachteristics (dimensions, primary/recurrent, uni/poly-lobulated) and its site of origin. Endoscopic approach has never been described as a feasible treatment for polyps presenting all the characteristics encountered in our experience [10,11]. *trans*-Cervical surgical resection should be reserved for complex cases, such as this case, in which a giant trilobular recurrent polyp had been unsuccessfully managed with endoscopic resection.

4. Summary

The presented case highlights the difficulties that can be encountered on managing a complex oesophageal polyp due to its dimensions, multiple implantation bases and its recurrent history. Different operative methods were engaged and eventually a cervicotomy excision resulted resolutive although a post-operatory fistula occurred.

As resulted from literature, giant oesophageal polyp management can be performed in several ways such as endoscopic, cervicotomy of thoracotomy or combining them, but there is no a clear indication of a gold standard procedure to adopt due to the rarity of case found.

By looking to our experience and to the previous cases reported that we analyzed, treatment choice has to be tailored on polyp's characteristics (dimensions, base and site of implantation, recurrence) and on patient's status (age, comorbidity, previous interventions).

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Contributors

Emilio Vicente and Yolanda Quijano proposed the study. Valentina Ferri and Riccado Caruso performed research and wrote the first draft. Hipolito Duran, Eduardo Diaz, and Isabel Fabra collected and analyzed the data. All authors contributed to the design and interpretation of the study and to further drafts. Valentina Ferri is the guarantor.

Ethical approval

This study was approved by the Ethics Committee of the Sanchinarro Hospital, San Pablo University.

This article was written following the SCARE Guidelines 2020: http://www.scareguideline.com [12].

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Declaration of competing interest

All the authors declare not to have any real or apparent conflicts of interest that may have a direct influence on the subject matter of the article.

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