



Case report

Giant fibrovascular polyps of the esophagus. Trans oral versus surgical approach. Case report and systematic literature review



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ABSTRACT

Introduction: Giant fibrovascular esophageal polyp is a rare benign intraluminal tumour. The aim of this study is to perform a review of the most recent literature in order to describe and analyse the current range of possible diagnostics and treatment strategies.

Case report: We present two cases of giant fibrovascular esophageal polyp treated with a combined minimally invasive transluminal approach at Sanchinarro University Hospital. Further, we perform a literature review.

Conclusion: We present two cases of giant fibrovascular polyp submitted to minimally invasive transluminal approach. Furthermore, 54 original articles reporting 59 cases have been analysed. In the surgical group, an esophagotomy and polyp resection were performed in 31 (91 %) patients and a total esophagectomy in two patients (5,8 %). Severe morbidity occurred in two patients (5,8 %). The median hospital stay was 9.25 days. A total of two (5,8 %) cases of recurrence have been registered. In the minimally invasive transluminal approach group, 27 patients had a polyp resection performed completely by endoscopy/transoral. There were no complications but there was one case of recurrence.

Conclusion: The transluminal approach is safe and should be considered also in the treatment of large esophageal polyps.

1. Introduction

Giant fibrovascular polyps of the esophagus (GFE) are rare, benign tumours arising from the cervical esophagus or hypopharynx. GFE are usually larger than 4 cm, although they can grow to a considerable size before becoming symptomatic [1]. The common symptoms include: dysphagia, regurgitation, odynophagia, neck pain, respiratory distress, or gastrointestinal bleeding [2]. The diagnostic and therapeutic management can be challenging and often requires multimodality radiologic, endoscopic and surgical strategies. The minimally invasive transluminal approach may be superior with respect to the conventional surgical approach and is progressively gaining acceptance. Though only a small number of cases have been reported in the literature, the aim of this paper is therefore to present our experience with GFE treated with transluminal approach, and to conduct a literature review that highlights the current range of strategies available in the treatment of GFE.

2. Materials and methods

We add to the current literature two cases treated at HM Sanchinarro University hospital with a minimally invasive transluminal approach. Written informed consent was obtained from patients for publication of these case reports and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. The work has been reported in line with the SCARE 2020 criteria [3].

A systematic search on the PubMed database using MESH terms: "Lipoma" OR "Polyps" OR "Fibroma" OR "Hamartoma" AND "Esophagus" OR "Esophageal Neoplasms" was performed. Original articles in human and adult published from 2004/01/01 to 2021/01/01 were included (Fig. 1). A literature search was performed by two independent researchers and only English language articles were considered.

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2.1. Data collection

The main patient demographic characteristics (age, sex, tumour size) were collected. Operative data were evaluated and the type of approach was used to divide patients into different groups (Fig. 2). Patients who submitted to open surgery carried out by cervicotomy or thoracotomy were included in the surgical group, while patients submitted to the transluminal approach were divided in two groups: first, the transoral group, where a surgical per-oral section of the polyp was obtained with the use of laryngoscope or a diverticuloscope plus stapler or electrical, surgical devices; and second, the endoscopic group, where the exeresis was conduct only with endoscopic instrumentation (Endoloop, snare).

Postoperative morbidity was stratified according to the Clavien–Dindo classification system [4], and severe morbidity was identified when grade \geq III occurred.

2.2. Statistical analysis

Continuous variables are reported as medium with standard deviation and categorical variables as absolute frequency and percentage. Variables are compared with the Wilcoxon rank-sum test and chi-square for quantitative and qualitative data, respectively.

3. Case presentation

3.1. Case 1

A 43-year-old male patient was referred to our service for episodes of regurgitation of a mass in the mouth. Family history and past medical history were uneventful. CT scan and MRI detected a 23 (Fig. 3) cm pedunculated lump filling the entire lumen of esophagus up to the gastric fundus arising from the cricoesophageal junction.

The echoendoscopy confirms the previous radiological findings. Furthermore, it showed a stalk of 2.5 cm diameter with a rich internal vascularization.

A combined trans oral/endoscopic resection was planned and performed under general anaesthesia with a nasotracheal intubation.

With a flexible esophagoscopy, the distal end of the polyp was gradually extracted through the oral cavity by an expert gastroenterologist (SP). An endo-Gia stapler was used to staple across the base of the attachment and two staple cartridges were fired removing the polyp. The esophageal lumen was then endoscopically checked and no bleeding, mucosal tears or perforation were found. The patient had an uneventful recovery and was discharged on the second postoperative day. The specimen showed a pathological benign fibrovascular proliferation confirming the preoperative diagnosis. An endoscopy performed

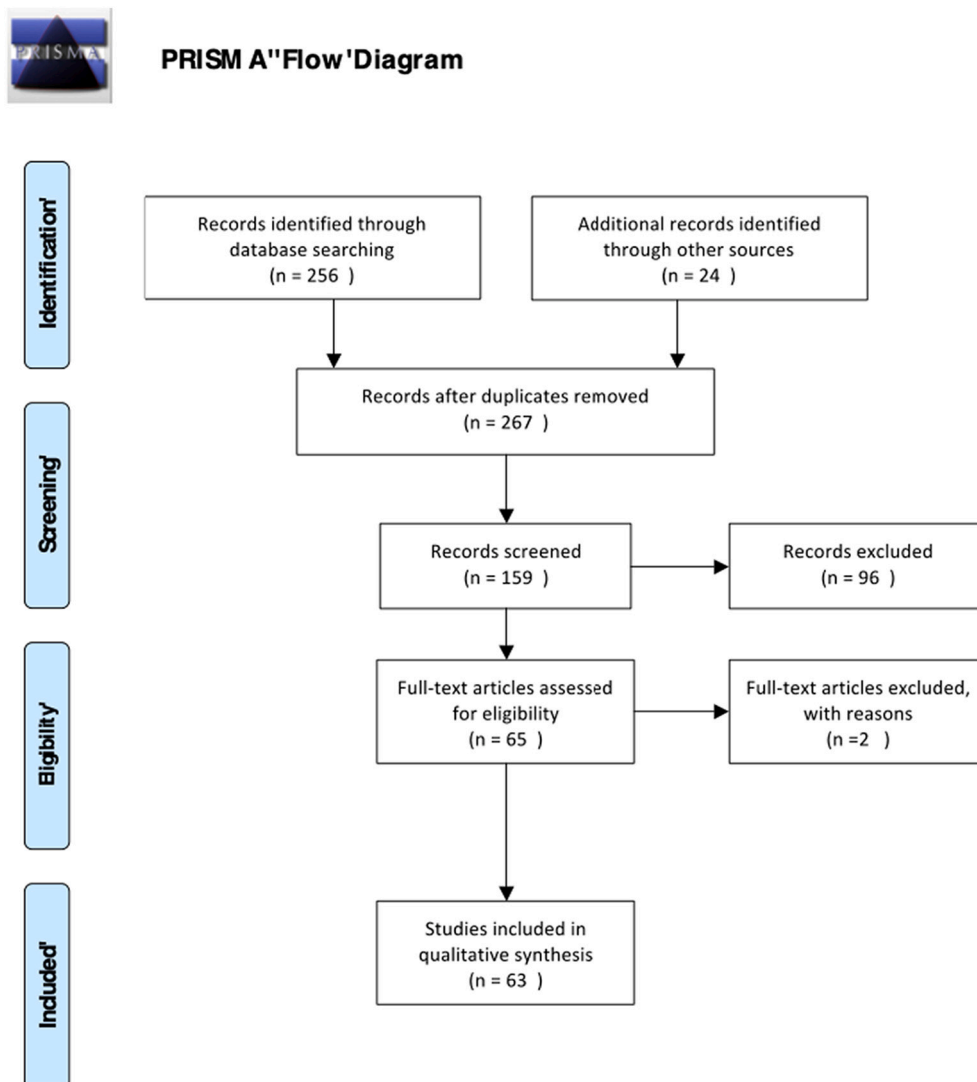


Fig. 1. PRISMA Flow chart. Studies selection.

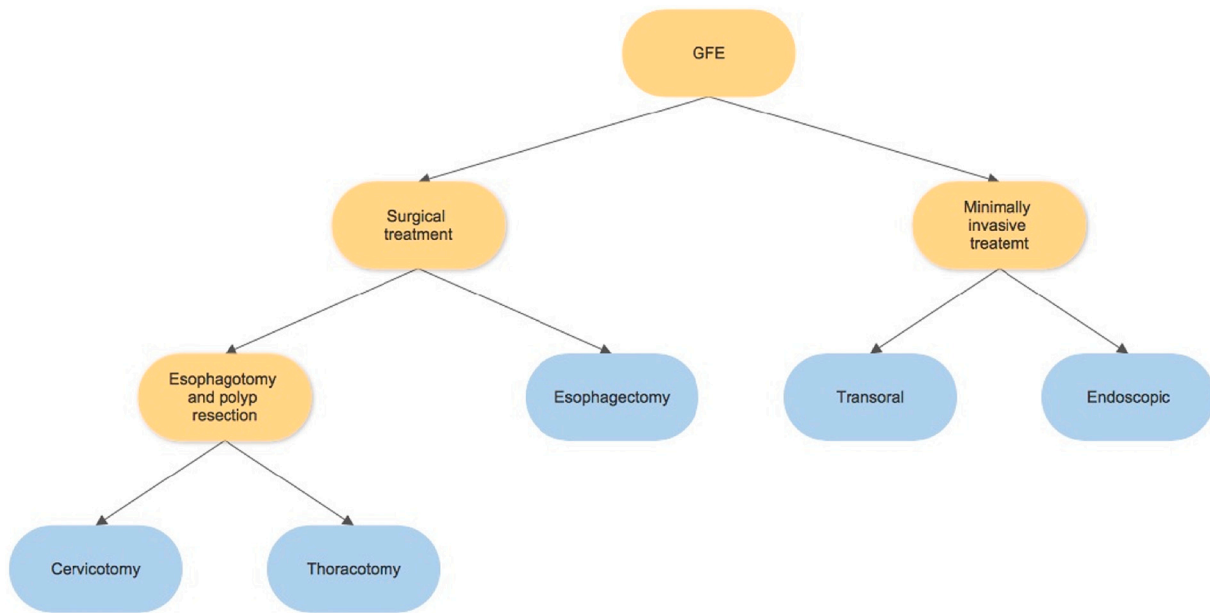


Fig. 2. Flow chart: different approaches in the treatment of GFE.

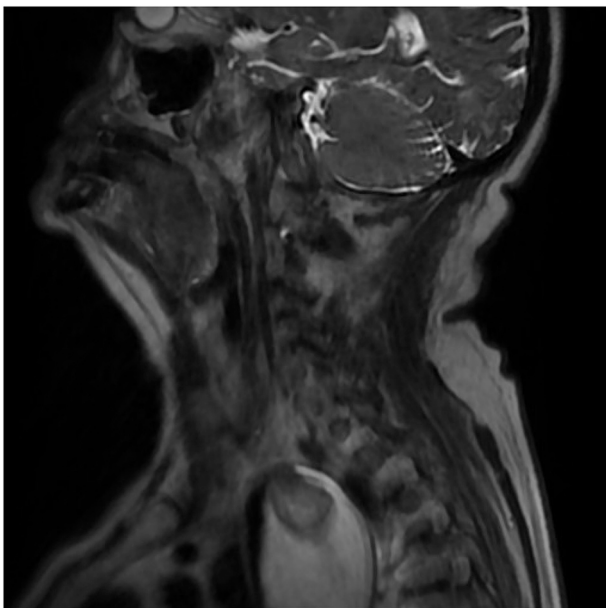


Fig. 3. MRI imaging of case 1.

three months after the operation was normal without recurrence at five years of follow up.

3.2. Case 2

A 74-year-old male patient presenting with a sudden dysphagia and odynophagia was found to have an esophageal mass of 4.5 cm in length arising from the cricopharyngeal junction on the CT scan. The patient underwent an MRI and endoscopy (Fig. 4) and endoscopic ultrasound, both confirming the presence of a giant polyp. Because of its short length, we used the Weerda diverticuloscope for removing the polyp through the oral cavity. The endo-Gia stapler was used to resect the polyp with the cartridge. The anatomic-pathologic examination confirmed a pathological benign fibrovascular proliferation.

The patient was discharged two days after surgery without any

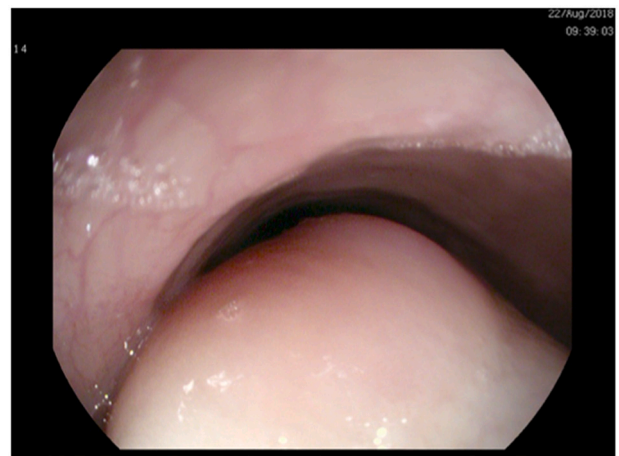


Fig. 4. Endoscopic view of the polyp in case 2.

complication. At three months of follow up, he remains asymptomatic.

3.3. Literature review

We identified 54 original articles that described 59 cases of GFE. Including our series, we considered a total of 61 patients in our analysis.

Table 1 Demographic data.

	Tot N 61	Transluminal group N 27	Surgical group N 34	p
Age (years ± SD)	56,42 (±12,58)	58,29 ± 12,80	54,97 ± 12,37	0,414
Sex M/F	44/17	19/8	25/9	0,369
Diameter (mm ± SD)	132,51 (±58,60)	114,32 ± 54,84	147 ± 57, 98	0,014
Hospital stay (days ± SD)	7,55 ± 9,25	6,42 ± 12,22	9,25 ± 5,17	0,488
Complication Clavien > 3	2	0	2	0,129
Recurrence	3	1	2	0,465

The median age was 56.42 years, 44 patients were male and 17 were female. The medial size of the lesion was 138.5 mm (Table 1): 147 mm in the surgical group and 114.32 mm in the transluminal group ($p < 0.014$). Symptoms are reported in Table 2.

In the surgical group (Table 3), access to the esophagus made through a lateral cervicotomy, followed by esophagectomy and polyp resection was performed in 31 (91 %) patients. Polyp resection via trans-thoracic approach was performed in two (5,8 %) patients. The combined approach (left cervicotomy, laparotomy and left thoracotomy) was required in one (2,9 %) patient. Two (5,8 %) patients were treated with a total esophagectomy. Polyp extraction through gastrotomy was performed in five (14,7 %) patients; two of which used the laparoscopic approach.

In the transluminal group, 27 patients have been analysed, 14 were treated with the transoral approach (Table 4) and 13 patients with the endoscopic approach (Table 5).

In the surgical group, three cases (8,8 %) of esophageal fistula have been described; two of which have been treated conservatively. Furthermore, a case of transient dysphagia and a case of pneumothorax have been reported. Severe morbidity occurred in three patients (11,1 %). In the transluminal group, two cases of transient dysphagia have been reported, while neither major complication nor postoperative mortality has been described.

Median hospital stay was 7.55 days, 9.25 days in the surgical group and 6.42 days in the transluminal group, without a statistical correlation ($p > 0.05$).

A total of two (5,8 %) cases of recurrence have been registered in the surgical group and one case (3,7 %) in the transluminal group ($p > 0.05$).

Two cases of recurrence have been reported in the surgical group and one case of recurrence has been described in the transluminal group ($p > 0.05$).

4. Discussion

According to World Health Organization classification, the umbrella term 'fibrovascular polyp' includes esophageal pedunculated benign tumours, such as fibroma, fibrolipoma, fibromyxoma or lipoma [1]. Length can vary from a few centimetres to up to almost 30 cm; the average polyp length was found to be 13.9 cm (range, 2 cm to 27 cm) in our series. The pedicle represents the narrower and the more vascularized part of the polyp.

Cases with a simultaneous presence of two polyps have been reported [6], and occasionally GFE can be multilobate and present with an ulceration on their distal portion, possibly due to contact with the acidic contents of the stomach. Microscopically this lesion is usually covered with a typical stratified squamous epithelium and presents a core of mature fibromyxoid tissue with the variable presence of fibrous or mature adipose tissue [7].

Pathogenesis is a matter for discussion. Some authors claim that GFE arises from the pharyngo-esophageal junction in the Laimer-

Haeckermann, where a lack of muscular support might cause a progressive elongation of tissue due to peristalsis traction and swallowing [5]. On the contrary, Yu et al. [8], who performed a cytogenetic study, support that GFE presents multiple complex chromosomal changes with signs of ring instability that could suggest that GFE is a neoplastic process rather than a consequence of redundant hamartomatous esophageal tissue. Graham et al. [9] retrospectively retrieved and reanalysed 13 cases of esophageal cases coded as 'giant fibrovascular polyp,' 'lipoma' and 'liposarcoma', and found MDM2 amplification in all cases, suggesting that the great majority of large polypoid fat-containing masses of the esophagus represent well and dedifferentiated liposarcoma.

In GFE, malignant transformation is a very rare: the lipomatous components can undergo sarcomatous changes and the squamous mucosa can develop into squamous carcinomas, as respectively reported by Valladium et al. [10] and Cockelaire et al. [11].

4.1. Clinical presentation

GFE can be totally asymptomatic and incidentally diagnosed, or in other patients can mimic different pathology as described by Ansaloni et al. [12], who reported a GFE misdiagnosed with a thyroid nodule. On the other hand, the onset of the disease can be dramatically fatal; in fact 10 cases of sudden death for asphyxia have been reported [13]. Due to the indolent nature and the potential space that the esophagus provides, GFE can grow to considerable sizes without causing many symptoms. Typically, patients with GFE present dysphagia and regurgitation of the mass. Respiratory symptoms, chest pain, weight loss, melena and hematemesis, vocal cord paresis and epigastric pain have been also described. Caceres et al. [2] reported that 62 % of the patients had dysphagia, 38 % had regurgitation of the mass, 25 % reported a persistent lump in the throat, 19 % reported weight loss, plus regurgitation of food (14 %), non-exertional chest pain (8 %), persistent cough (7 %), odynophagia (7 %), sore throat (5 %), vomiting (2 %), abdominal pain (1 %), and melena (1 %).

4.2. Diagnosis

The diagnostic challenge consists of distinguishing between an intramural or an intraluminal mass. The diagnostic process should include a thorough history and a physical examination, followed by an upper endoscopy and barium or Gastrografin swallow.

When a GFE is suspected, EUS can provide information on the size, origin of the stalk and vascularity of the polyp. In addition, EUS-fine needle aspiration may add a histological sample. However, it remains that computed tomographic (CT) scan and magnetic resonance imaging (MRI) are considered the gold standard in determining the nature and origin of the mass. In 18 FDG/PET-TC imaging, GFE can present a pathologic FDG capitation [14].

4.3. Treatment

Considering the potentially fatal complications, excision of GFE is mandatory. This can be accomplished by surgical or by transluminal approach. Regarding the anaesthesiological preparation, in some patients the need to ensure the airway by means of tracheostomy has been described [15].

4.3.1. Surgical treatment

The surgical approach has represented the standard technique in GFE management until the establishment of endoscopic technology. The surgical approach is mainly recommended for large polyps [2], because of the risk of uncontrolled bleeding during stalk section. Over the last 30 years, we have found in our literature review a total of 40 patients who were submitted to surgery. The median size of the GFE treated with surgery was bigger with respect to the transluminal group with a statistical significance. Esophagotomy and polyp resection remains the

Table 2
Clinical presentation.

Symptoms n (%)	Tot N 61	Transluminal group N 25	Surgical group N 36
Dysphagia	37	15	22
Regurgitation	17	10	7
Chest pain	1	0	1
Weight loss	11	2	9
Melena/hematemesis	3	0	3
Vocal cord paralysis	1	0	1
Respiratory symptoms	15	10	5
Epigastric pain	2	2	0
Heartburn	3	2	1
Odinophagia	4	2	2

Table 3
Surgical group.

Author	n	Sex M/ F	Age	Size mm	Approach	Surgical technique	Complication	Recurrence (%)
Pinto 2018	1	M	23	200	Left cervicotomy	Esophagotomy, polyp resection	No	Nr
Cockbain 2017	3	2/1	42, 55, 56	150	Left cervicotomy, VL gastrotomy (E)	Esophagotomy, polyp resection	No	33 %
Cano 2017	1	M	60	200	Cervicotomy, gastrotomy (E)	Esophagotomy, polyp resection	No	No
Ongkasuwan 2016	2	1/1	63	50, 62	Lateral pharyngotomy	Esophagotomy, polyp resection	No	No
Sestini 2016	1	M	70	130	Right cervicotomy	Esophagotomy, polyp resection	No	Nr
Ansaloni 2016	1	W	54	200	Cervicotomy	Pharyngotomy, polyp resection	Esophageal fistula	No
Qinying 2015	1	M	52	45	Right cervicotomy	Esophagotomy, polyp resection	No	Nr
Zhang 2015	1	M	59	140	Left cervicotomy	Esophagotomy, polyp resection	No	No
Madeira 2013	1	M	47	230	Thoracotomy, laparotomy	Transthoracic esophagectomy	Pneumothorax	No
Cordos 2012	1	W	59	270	Cervicotomy	Esophagotomy, polyp resection	No	No
García et al. 2012	1	M	58	160	Left cervicotomy	Esophagotomy, polyp resection	No	No
Yu 2012	1	W	49	180	Cervicotomy + gastrotomy (E)	Pharyngotomy + esophagotomy polyp resection	No	No
Sweeney 2011	1	W	64	160	Left cervicotomy	Esophagotomy, polyp resection	No	No
Goenka 2011	1	M	47	170	Cervicotomy	Esophagotomy, polyp resection	Esophageal fistula	No
Jose 2010	1	M	55	200	Left cervicotomy, VL gastrotomy (E)	Esophagotomy, polyp resection	No	Nr
Ubukata 2010	1	M	74		Cervicotomy	Esophagotomy, polyp resection	No	Nr
Peltz 2010	1	M	79	130	Left cervicotomy	Esophagotomy, polyp resection	No	Nr
George 2009	1	M	52	200	Cervicotomy	Esophagotomy, polyp resection	No	No
Lee 2009	1	M	61	60	Left cervicotomy, laparotomy left thoracotomy	Esophagotomy, polyp resection	No	100 %
Dutta 2009	1	W	25	150	Cervicotomy tracheostomy	Esophagotomy, polyp resection	Esophageal fistula	No
Been 2009	1	M	54	50	Left cervicotomy	Esophagotomy, polyp resection	No	No
Liu 2008	1	M	67	110	Right thoracotomy	Esophagotomy, polyp resection	No	Nr
Blacha 2008	1	M	73	100	Left cervicotomy	Esophagotomy, polyp resection	Transient dysphagia	Nr
Kanaan 2007	1	W	60	180	Cervicotomy, laparotomy	Trans-hiatal esophagectomy	No	No
Luthen 2006	1	W	52	135	Right cervicotomy	Esophagotomy, polyp resection	No	Nr
Ridge 2006	1	M	42	Nr	Nr	Nr	No	Nr
Sultan 2005	2	1/1	38–58	160	1. Right cervicotomy 2. Right postero-lateral thoracotomy	Esophagotomy, polyp resection	No	No
Solerio 2005	1	M	74	180	Left cervicotomy	Esophagotomy, polyp resection	Nr	Nr
Kim et al. 2005	1	M	63	255	Cervicotomy + gastrotomy (E)	Esophagotomy, polyp resection	Nr	Nr
Ozcelik 2004	1	W	51	100	Left cervicotomy	Esophagotomy, polyp resection	No	Nr

Table 4
Transoral group.

Author	n	Sex	Age	Tumour size, mm	Access	Resection technique	Complication	Recurrence (%)
Present study	2	M	43, 74	230, 45	Weerda laryngoscope	Endoscopic guide extraction EndoGIA	No	No
Mana 2019	1	M	42	50	Rigid esophagoscope Dilating laryngoscope	Endo-Gia	No	Nr
Lobo 2016	1	W	58	70	Pharyngo-scope with suspension arm	Harmonic scalpel	No	Nr
Hinton-Bayre 2016	1	M	55	160	Weerda laryngoscope	Endoloop, endo-Gia	No	No
Valiuddin 2016	1	M	68	130	Weerda laryngoscope	Snare	No	No
Liu 2014	1	M	50	60	Transoral	Electric coagulation	No	Nr
Włodarczyk 2013	1	M	54	139	Transoral resection	Electric coagulation	No	Nr
Kau 2012	1	W	38	150	Weerda laryngoscope Microscope	CO2 laser	No	No
Ozdemir 2011	1	M	44	110	Mouth gag and retractor. Tracheo	Electric coagulation	No	No
Goto et al., 2010	1	M	45	70	Transoral resection		No	Nr
Ivan et al., 2009	1	M	62	100	Weerda laryngoscope Microscope	Snare + ligation + electric coagulation	No	Nr
Fumagalli 2008	1	W	54	200	Weerda diverticuloscope	Laparoscopic scissors	No	Nr
Pham 2008	1	M	63	50	Weerda laryngoscope	Bipolar cautery, snare	No	Nr

gold standard approach and lateral cervicotomy is the most commonly used method of access. Polyp resections via trans-thoracic approach have been performed in only two patients. In the first case described by Liu et al. [16], the trans-thoracic approach was preferred for size and location of GFE and for the risks of airway compression. Moreover, a combined approach (cervicotomy/laparotomy/thoracotomy) has been described due to the impossibility in retracting the polyp cranially

neither through cervicotomy nor through thoracotomy [17]. Due to a suspected diagnosis of an intramural leiomyoma and a GIST, a total esophagectomy was performed in two patients [18]. With a larger polyp, if extraction through cervicotomy is not feasible, gastrotomy can be performed, also with the laparoscopic approach.

Table 5

Endoscopic group.

Author	n	Sex	Age	Tumour size, mm	Access	Resection technique	Complication	Recurrence (%)
Fedorov 2018	1	W	52	135	Double-channel gastroscope	Endoloop, snare	No	Nr
Ward 2016	1	M	62	160	Gastroscope	Snare	No	Nr
Cockbain 2017	1	M	72	110	Gastroscope, VL gastrotomy (E)	Needle-knife	No	No
Jo 2016	1	W	45	100	Gastroscope	Snare	No	Nr
Li 2016	1	M	50	180	Gastroscope	Snare	No	Nr
Ongkasuwan 2016	2	2 M	77, 85	25, 170	Gastroscope	Bovie cautery	No	50 %
Lorenzo 2016	1	W	66	150	Large-channel gastroscope	Electrosurgical knife	No	Nr
Fernandes 2015	1	M	55	150	Gastroscope	Endoloop, snare	No	Nr
Di Mitri 2014	1	W	51	200	Operative, single channel endoscope	Snare, electric coagulation	No	Residual stalk
Murino 2014	1	M	50	90	Adult gastroscope, paediatric gastroscope	Endoloop, snare	No	Nr
Chauhan et al., 2011	1	W	81	120, 100	Double-channel upper endoscope	Snare	No	Nr
Alobid 2007	1	M	76	100	Flexible esophagoscopy	Endoloop + snare	No	Nr

4.3.2. Minimally invasive transluminal treatment

GFE can be amenable to transluminal resection; indeed, the peduncle of the polyp does not contain the deeper muscular layer of the esophagus, and endoscopic resection can be proposed with very limited risk of perforation. Minimally invasive transluminal treatment may not yet be well known by surgeons, and for this reason, our review analyses the key points of this approach in detail.

In our review, we found 31 cases of patients who were submitted to transluminal resection. In 15 cases, a transoral resection was performed. The Weerda laryngoscope and the Weerda diverticuloscope (Karl Storz Endoskopie GmbH, Tuttlingen Germany) were the most frequently used instruments. The Weerda laryngoscope can be used in conjunction with the operating microscope to facilitate incision through the stalk. Ivan et al. [19] 2008 believe that in the case of a giant fibrovascular polyp, transoral resection is a safe approach if: “(1) it can be reached with the Weerda laryngoscope, (2) the origin of the polyp can be well visualized, (3) the polyp has a stalk, (4) the suture ligation of the stalk can be used safely, and (5) over the suture ligation the polyp can be easily resected”.

The endoscopic removal of GFE is more challenging because of the difficulty of trapping the polyp stalk, mostly with the larger size polyps. This minimally invasive approach has been gaining relevance in recent years, thanks to the development of new and more flexible endoscopic guide that allow both the extraction of the polyp and its section with direct vision.

The technical devices used for endoscopy are different, and the correct approach to these large polyps should be decided on a case-by-case basis. The most utilized is Endoloop, which can be put in the polyp stalk and sectioned with an electrosurgical snare. The polyp can then be retrieved trans-orally or through gastrotomy.

In the first report of our case series, a combined endoscopic/transoral approach was carried out. A flexible esophagoscopy of the distal end of the polyp was gradually extracted through the oral cavity and then an endo-Gia stapler was used for the stalk section. In our opinion this is a safe option for large polyp, in order to achieve a better control of the polyp stump.

In this study, we report an extensive review in the treatments for GFE over the last 30 years, the first such reported literature review to now. Nevertheless, some limitations are present; primarily in the absence in many reports of data concerning hospital stay, complication and recurrence.

In our review, we have observed that no major complication was observed in the minimally invasive transluminal group, while in the surgical group three esophageal fistula, a transient dysphagia and a pneumothorax have been reported. A case of postoperative death has been described in the surgical group in a patient with a squamous carcinoma that originated from the polyp with advanced lympho-node metastatic disease. Hospital stay was inferior in the endoscopic group, with respect to the surgical one, but without a statistical correlation. Few reports have provided follow up data; we identified three cases of recurrence in the surgical group and two cases in the transluminal group without statistical significance.

Finally, the minimally invasive transluminal approach should represent the gold standard in the treatment of cases of giant polyps and the multidisciplinary management of GFE may lead to a successful and safe control of the stalk. In fact, previous extractions of the entire polyp through the mouth using an endoscopic guide, as reported for the first time in our patient, can assure better control of the stalk during transoral surgical resection.

5. Conclusion

The minimally invasive transluminal approach represents a safe and feasible option in the treatment of giant fibrovascular polyps of the esophagus, compared with the surgical approach, showing similar rates of recurrence and absence of major complications.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Consent

Written informed consent was obtained from patients for publication of these case reports and accompanying images.

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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Research registration number

None.

CRediT authorship contribution statement

Emilio Vicente and Yolanda Quijano proposed the study. Valentina Ferri performed research and wrote the first draft. Susanna Prados, Roberta Isernia y Riccardo Caruso collected and analysed the data. All authors contributed to the design and interpretation of the study and to further drafts.

Declaration of competing interest

Authors declare no conflict of interest.

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