



Giant esophageal fibrovascular polyp with clinical behaviour of inflammatory pseudotumor: A case report and the literature review

Džinovski fibrovaskularni polip jednjaka sa kliničkim ponašanjem inflamatornog pseudotumora

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Abstract

Introduction. Esophageal fibrovascular polyps are rare, benign, intraluminal, submucosal tumor-like lesions, characterized by pedunculated masses which can demonstrate enormous growth. The most frequent symptoms are dysphagia, vomiting and weight loss. Fibrovascular polyps with long stalks can regurgitate into the airways and cause asphyxia. Esophageal inflammatory pseudotumor is extremely rare lesion accompanied with various systemic manifestations as fever, anemia and thrombocytosis. **Case report.** We presented a 29-year-old man complaining of a long-lasting fever and dysphagia. He was found to have huge pedunculated submucosal tumor of esophagus, surgically completely resected. Histopathological examination showed that this giant tumor, 24 × 9 × 6 cm, was a fibrovascular polyp. The postoperative course was uneventful. The preoperative fever, anemia and thrombocytosis disappeared and did not recur in the postoperative course. **Conclusion.** We reported a patient with giant esophageal pedunculated tumor with clinical manifestations of inflammatory pseudotumor and histopathological picture of fibrovascular polyp, that we have not found described in the literature before.

Key words:

polyps; esophagus; granuloma, plasma cell; diagnosis; endosonography; histological techniques; surgical procedures, operative; treatment outcome.

Apstrakt

Uvod. Fibrovaskularni polipi jednjaka su retke, benigne, intraluminalne, submukozne neoplazme na peteljci, koje mogu postići džinovski rast. Najčešći simptomi fibrovaskularnih polipa jednjaka su otežano gutanje, povraćanje i mršavljenje. Fibrovaskularni polipi sa dugom peteljkom se mogu povratiti u disajne puteve izazivajući asfiksiju. Inflatorni pseudotumor jednjaka je veoma redak tumor, često udružen sa različitim sistemskim manifestacijama kao što su febrilnost, anemija i trombocitoza. **Prikaz bolesnika.** Prikazali smo bolesnika starog 29 godina, sa dugotrajnom febrilnošću i disfagijom. Tokom ispitivanja kod bolesnika je nađen veliki submukozni tumor jednjaka na peteljci, koji je hirurški odstranjen u celosti. Histopatološki pregled ovog velikog tumora, 24 × 9 × 6 cm, pokazao je da se radilo o fibrovaskularnom polipu. Postoperativni tok je protekao bez komplikacija, a preoperativna febrilnost, anemija i trombocitoza su nestale. **Zaključak.** Ovo je prikaz slučaja bolesnika sa velikim polipoidnim tumorom jednjaka koji se klinički ponašao kao zapaljenski pseudotumor, a mikroskopska slika je odgovarala fibrovaskularnom polipu, što nismo našli opisano u literaturi do sada.

Ključne reči:

polipi; jednjak; granulom, plazmocelularni; dijagnoza; endosonografija; histološke tehnike; hirurgija, operative procedure; lečenje, ishod.

Introduction

Fibrovascular polyps (FPs) are rare, benign, intraluminal, submucosal, tumor-like lesions characterized by the development of pedunculated, intraluminal masses, which in

the esophagus can demonstrate an enormous growth. Dysphagia, vomiting, weight loss, and respiratory symptoms are the most frequent complaints of patients with FP, and these with long stalks can regurgitate into the pharynx or mouth and cause asphyxia¹.

Inflammatory pseudotumors (IPTs) are benign and rare lesions, forming a group of etiologically, histologically, and biologically heterogeneous lesions that are histologically characterized by prominent inflammatory infiltrates². IPT has been described in various organs but esophageal localization is extremely rare. These quasineoplastic lesions may mimic a malignant tumor clinically and radiologically³. Clinical presentation of patients with IPTs tends to be with varying degrees of fever, iron-refractory anemia, and thrombocytosis⁴.

We presented a 29-year-old male with fever of unknown origin, malaise, anemia and thrombocytosis, who was diagnosed and operated for a really giant esophageal polypoid tumor. The clinical manifestations of this unique lesion with behavior of IP and histopathological picture of FP were described with a review of the literature.

Case report

A 29-years-old Caucasian male, without any previous medical history, was referred to the Military Medical Academy (MMA) Belgrade, Serbia, for further investigation due to throat pain, dysphagia, and fever up to 39°C, malaise, anemia and thrombocytosis, present for few months. The patient had nonselective anorexia and lost 7 kg of his body mass for the last 4 months.

In the moment of the patient referral to MMA, his status during physical examination was normal. There was no organ enlargement, ascites or edema. Body temperature showed high fever, up to 39°C.

Laboratory investigations showed some increased values: the erythrocyte sedimentation rate (ESR) was 98 mm/h (normal: < 15 mm/h), C-reactive protein (CRP) 16.72 mg/L (normal range 0–3 mg/L), fibrinogen 8.9 g/L (normal range 2–4 g/L) platelets $833 \times 10^9/L$ (normal range $140\text{--}450 \times 10^9/L$) and gamma-glutamyl transferase (GGT) 78 U/L (normal range 0–38 U/L). There was a decreases in the levels of red blood cell (RBC) $3.6 \times 10^{12}/L$ (normal range $3.9\text{--}5.7 \times 10^{12}/L$, hemoglobin (Hgb) 83 g/L (normal range 120–180 g/L) and hematocrite (Hct) 0.27 L/L (normal range 0.36–0.503 L/L). A blood film showed a microcytic, hypochromic picture with hypersegmented neutrophils. Urine sample was normal and repeated blood, urine and stool cultures were negative. There was no serological evidence of infection due to cytomegalovirus (CMV), Epstein-Barr virus (EBV), adenovirus, brucellosis, psittacosis, mycoplasma, legionella, toxoplasma or Q fever. Thyroid hormones were within normal range. Bone marrow smear showed reactive hyperplasia. The purified protein derivative (PPD) skin test was negative. Rheumatoid factors and antinuclear antibodies (ANA) were negative. Serum immunoglobulins were within normal limits.

Electrocardiogram (ECG) and echocardiogram showed a normal heart function without evidence of vegetations. Chest x-ray was normal as abdominal echosonogram.

Initial upper endoscopy demonstrated a large pedunculated mass starting from cervical part of esophagus, on 21st cm from the front teeth there was a stalk with 3 cm in diameter, originating from the right esophageal wall. The tu-

mor stretched across the entire esophagus, up to gastric cardia. Tumor was covered with overlying mucosa and had mucosal erosion on the top, without actual bleeding. The stomach and duodenum were without any pathological changes on mucosa. Initial biopsies were inconclusive.

Endoultrasonography (EUS) confirmed endoscopic findings. Pedunculated submucosal tumor was starting from lamina muscularis propria of cervical esophagus, the upper diameter was 3 cm and the distal 6–9 cm. The tumor was echoheterogenic, dominantly hypoechogetic.

Double-contrast barium meal examination demonstrated normal deglutition act and giant pedunculated intraluminal mass, which completely filled the esophagus, up to the gastric cardia, causing significant dilatation and obstruction (Figure 1). Tumor starts in the cervical part of esophagus and contrast normally passes in the stomach (Figure 2A and B).



Fig. 1 – Barium meal examination shows esophageal enlargement with a huge tumor inside.

A computed tomographic (CT) scan of the chest showed an intraluminal esophageal mass of $21 \times 9 \times 6$ cm, arising from the cervical part of the esophagus and reaching the cardiac orifice of the stomach. Tumor mass fulfilled and dilate the complete esophagus but without wall infiltration. There was no evidence of metastasis or lymphadenopathy in the thorax and abdomen (Figure 3).

After investigation was completed, operative treatment was suggested to the patient and he accepted it.

In general anesthesia we reached the esophagus through the left lateral neck incision. A left esophageal wall was incised in the length of 4 cm. Through this incision a huge pedunculated tumor with smooth wall was visible. The stalk of this tumor was 3 cm in its basis, originating from the right esophageal wall. The stalk was cut and the basis checked by frozen section, and no malignancy was found. The basis of the stalk was sutured by absorbable sutures. An attempt to remove the tumor through esophageal incision failed due to its huge caliber and the esophagotomy was sutured in a two-layered fashion. Median laparotomy was done and exploration did not show any pathological findings in abdomen. After

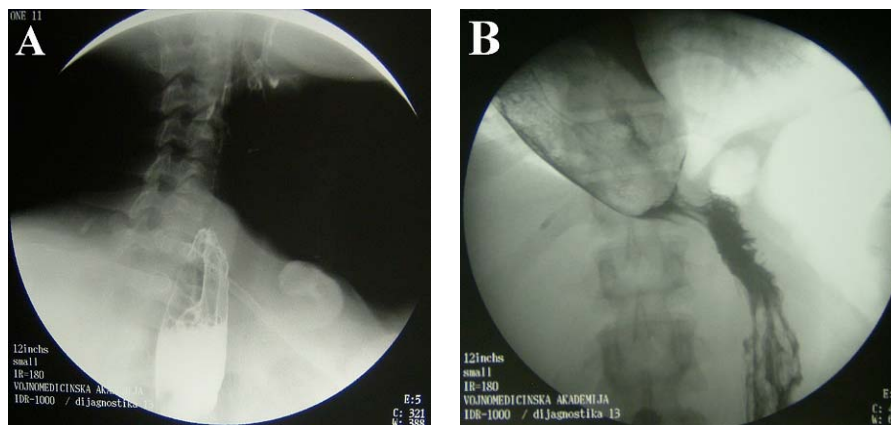


Fig. 2 – Barium meal examination: A) The tumor basis starts in the cervical esophagus; B) The top of the tumor is in front of the gastric cardia.



Fig. 3 – Thorax computed tomography – tumor fulfills the esophagus without wall infiltration.

gastrotomy we found no pathological content in the stomach. The tumor apex was prominent through the gastric cardia and the tumor was completely removed, “delivered”, through gastric incision (Figure 4). Gastrotomy was closed by a layer of running absorbable sutures, abdominal cavity drained with rubber drain and laparotomy closed.

Gross pathology demonstrated that the removed polypoid tumor was penile – like in shape, 21 × 9 × 6 cm (Figure 5). The surface of the tumor was mainly smooth except on the top where mucosal erosion was present (Figure 6). It was open longitudinally; tissue on the cutting surface was homogeneous, white color, smooth and shiny with foci of mixoid degeneration and with tough and elastic consistency (Figure 7). The proximal part of polyp ended with the stalk, 2 cm long.

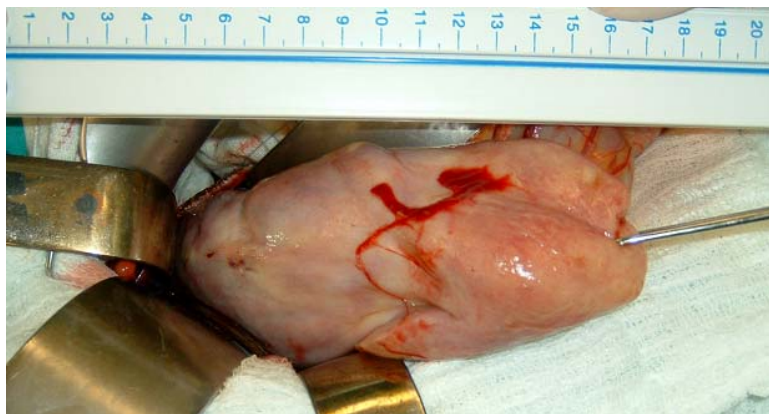


Fig. 4 – The tumor is “delivering” from the stomach.



Fig. 5 – A complete tumor with a stalk.



Fig. 6 – The tumor from the other side with mucosal ulceration.

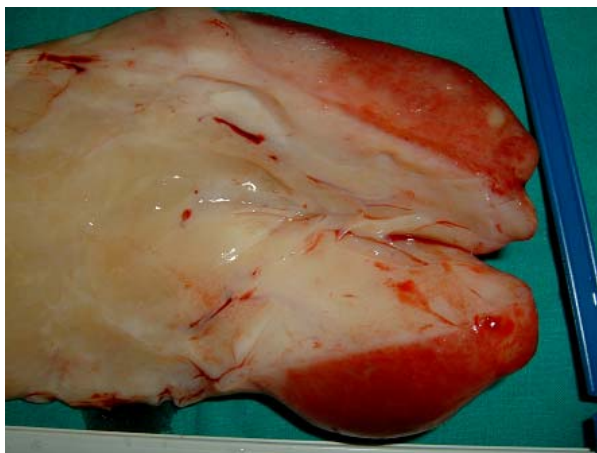


Fig. 7 – Cutting surface of the tumor.

Histologic sections were fixed in formalin, routinely processed, embedded in paraffin and stained with hematoxylin-eosin (H E). The immunohistochemistry analysis was routinely processed on formalin fixed paraffin embedded (FFPE) tissue sections thickness of 4 μ . After deparaffinization and blocking of endogenous peroxidase activity by 3% hydrogen peroxide, the sections were incubated with primary antibodies with a Dako EnVision system. The following antibodies were used: smooth muscle actin (SMA), CD34, CD68, Ki-67 and S-100 protein. After incubation with secondary antibodies, the sections were visualized with 3-3'-diaminobenzidine and were counterstained with Mayer's hematoxylin.

Microscopically, the polyp was mainly covered with mature squamous epithelium, except on the top (Figure 8A and B), where mucosal erosion was present and covered by necrotic detritus with underlying granulation tissue with diffuse infiltration of mixture of lymphocytes, plasma cells and neutrophils. In the base of granulation tissue few scattered atypical fibroblasts were found, mitotically inactive and seeming regenerative. The tumor was composed from the loose, focally mixoid and edematous collagen connective tissue with foci of hyalinization. Inside the tumor lobules of mature adipose tissue without lipoblasts were found (Figure 8C). The polyp was very well vascularized (Figure 8D and E) with multiplied blood vessels of small and medium caliber, venous and arterial type. Foci of inflammatory infiltrates, mainly lymphocytes and plasma cells, were present

not only around ulceration but also throughout entire polyp, especially perivascularly. Cappillary and bigger blood vessels showed a positive reaction of endothelial cells on CD34 (Figure 8F) and SMA, S-100 protein was negative. Inside the tumor there were histiocytes with immunoreactivity on CD68. In rare spindle cells Ki-67 showed positive reaction.

The postoperative course was uneventful. A double-contrast barium meal examination on 7th postoperative day showed no extravasation and normal passage through the esophagus and stomach and the patient started oral feeding. Postoperatively, the patient's body temperature was normal, anemia was corrected well as thrombocytosis (value of RBC was $4.1 \times 10^{12}/L$, Hgb 105 g/L, Hct 0.322% L/L, platelets $440 \times 10^9/L$).

Follow-up within 5 years showed that the patient was without any complaints, without dysphagia and fever, with laboratory findings in the normal range. Control upper endoscopy showed normal status on esophagus and stomach and there was no tumor recurrence.

Discussion

Fibrovascular polyps of the esophagus are rare benign tumors, comprising about 1% of all benign esophageal tumors. However, they are the most common submucosal tumor-like lesions of the esophagus, characterized by the development of pedunculated, intraluminal masses that, in the esophagus can demonstrate enormous growth. Giant FPs are defined as polyps larger than 5 cm in maximal diameter. They are slow growing, pedunculated tumor masses that often arise from the upper esophagus, at the pharyngo-esophageal junction, Laimer's triangle⁵.

Inflammatory pseudotumors are benign and rare lesions, forming a group of etiologically, histologically, and biologically heterogeneous tumefactive lesions that are histologically characterized by prominent inflammatory infiltrates⁴. IPT has been described in various organs, most commonly involving the lungs and orbit, but also found in nearly every site in the body⁶. These lesions may mimic a malignant tumor clinically and radiologically⁷. Recently there have been reports about multicentric localization of IPT⁸. Tumor size of IPT varies and can be very huge. Esophageal localization is very rare as their size over 20 cm⁹.

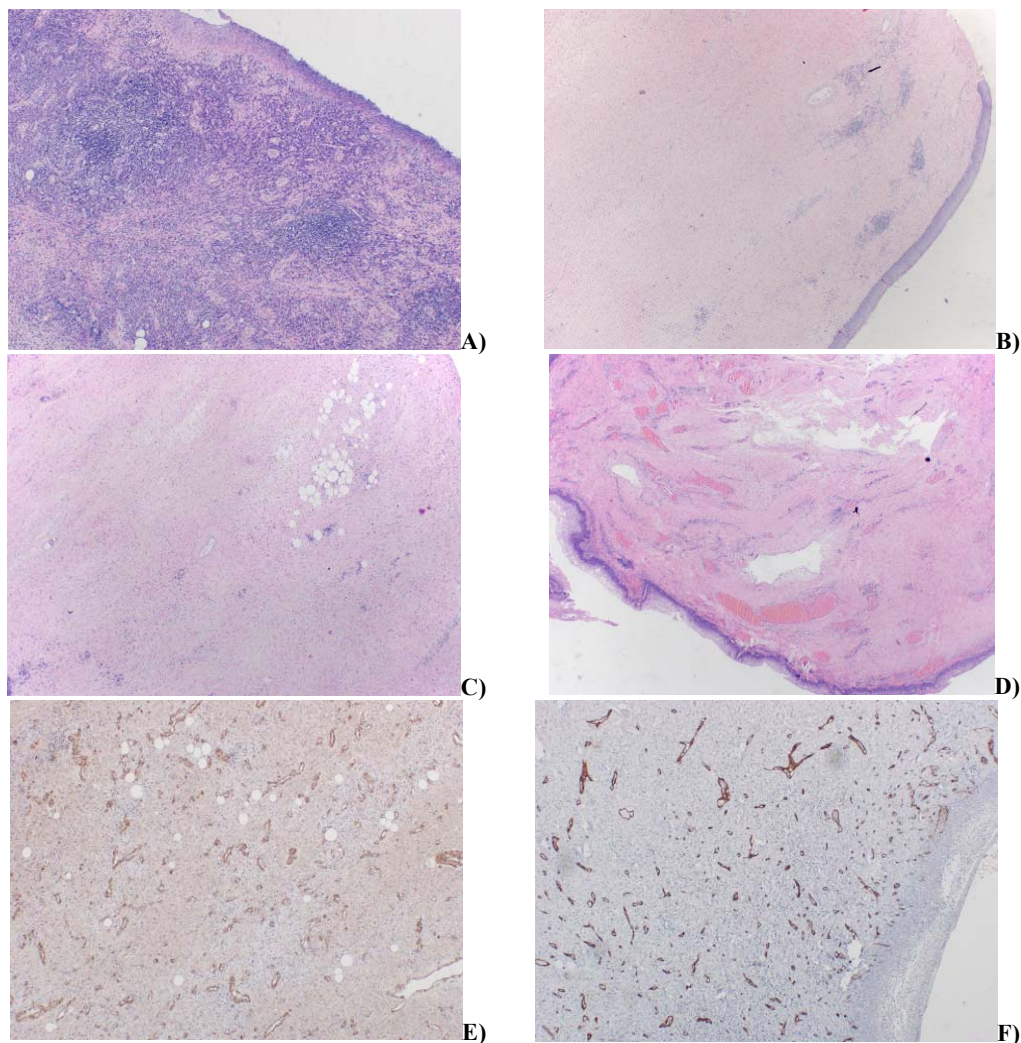


Fig. 8 – A) Mucosal erosion on the top of the polyp with the underlying granulation tissue infiltrated with lymphocytes, plasma cells, neutrophil and eosinophyl granulocytes (HE, $\times 20$); B) Regularly stratified squamous epithelium covering surface of the polyp out of ulceration (HE, $\times 4$); C) Fat tissue lobulus in the connective tissue (HE, $\times 10$); D) Multiplied blood vessels, small and medium calibar (HE, $\times 10$); E) Vascular spaces collored by actin (immunohistochemical reaction, LSAB+, $\times 10$); F) Endothelial cells with positive reaction on CD34 (immunohistochemical reaction, LSAB+, $\times 10$).

The majority of FPs occur in elderly people, aged 60–70 years, but they have been reported also in a 5 months old infant¹⁰. Although IPT can occur at any age, in both genders, it is most commonly present in children and young adults¹¹.

The pathogenetic origin of FP is from the loose and redundant submucosal tissue near the Laimer's triangle. Due to the lack of muscular support, this relatively mobile tissue through years of esophageal peristalsis, traction and swallowing, is dragged along, elongated and enlarged intraluminally¹². The etiology and pathogenesis of IPT remain unclear, reactive-infectious, immunological and idiopathic factors might play a role in their initiation and growth¹³. Speculated etiology includes viral infection, focal parenchymal necrosis with hemorrhage secondary to trauma, coagulopathy or surgical trauma¹⁴. An immune-autoimmune mechanism has also been implicated¹⁵.

Fibrovascular polyps are covered with normal mucosa and containing different amounts of fibrous, vascular, and adipose tissue. Based on their histological composition, these polypoid lesions have been termed as lipomas, fibromas, fibrolipomas, or fibroepithelial polyps in the literature. More recently, the World Health Organization has classified them as FPs, in their international histological classification system¹⁶. 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 can develop into adenocarcinoma¹⁷.

Inflammatory pseudotumors can present as a single mass or multiple masses with polymorphous inflammatory cell infiltrates and variable amounts of fibrosis, necrosis, granulomatous reaction, and myofibroblastic spindle cells. The term IPT denotes a histologically similar group of tumors, characterized by a spindle cell proliferation with a fibroin-

inflammatory appearance that has been reported under a variety of additional descriptive terms, such as atypical fibromyxoid nodule, inflammatory fibroid polyp, inflammatory pseudotumor, plasma cell granuloma, and pseudosarcomatous myofibroblastic proliferation. The gross features of esophageal IPT vary from polypoid to diffuse non-polypoid, and are likely to occur in the distal esophagus or the esophago-cardial junction¹⁸.

Since polyp is slowly growing, it may remain asymptomatic for years until it reaches a large size. Dysphagia, vomiting, chronic gastrointestinal bleeding, weight loss and respiratory symptoms are the most frequent complaints of patients with FPs. Though biologically benign, these giant FPs can have dramatic and even life-threatening presentations because these polyps with long stalks, however, can regurgitate into the pharynx or mouth and cause death from asphyxiation if the larynx is occluded¹⁹.

A distinguishing feature of IPT, in up to 50% of cases, is the presence of a varying degree of inflammatory syndrome consisting of persistent fever, weight loss, malaise, iron-refractory anemia, moderate leukocytosis, thrombocytosis, polyclonal hyper-globulinemia and elevated erythrocyte sedimentation rate. Many of these features can be related to the production of inflammatory mediators such as cytokines and particularly Interleukin-1, which has a wide range of local and systemic effects, as tumor-specific inflammatory response¹⁰. Our patient demonstrated several aspects of this syndrome, i.e. fever, malaise, weight loss, and thrombocytosis. Both clinical and laboratory manifestations tend to resolve rapidly after surgery⁴, as was the case in the presented patient.

Usually, the diagnosis is made by imaging and endoscopic studies. Barium double-contrast examination of the esophagus usually shows a sausage-shaped mass with multiple filling defects, which originates in the cervical esophagus and extends to the lower esophagus²⁰.

Endoscopy usually shows an intraluminal mass that is mobile and covered with normal mucosa. The presence of easily bleeding ulcer on the top can be observed by endoscopy, leading to the suspicion of malignancy. Careful examination of the upper esophageal sphincter may reveal the stalk of pedunculated mass. EUS has been reported as a method to demonstrate the submucosal origin of polyps²¹. EUS also provides information on a diameter of a polyp, as well as its vascularity at insertion point²². The submucosal location can make endoscopic or tru-cut biopsies difficult to obtain good specimens and this histopathological specimens are often inconclusive or misdiagnosed. The definitive diagnosis is often made based on histopathological analysis of surgically removed specimens of FP or IPT²³.

CT scanning and magnetic resonance imaging (MRI) can be useful to diagnose FP. In particular, MRI of the neck and thorax might be decisive in the choice of treatment by demonstrating the origin of the pedicle and the composition of a polyp. If the mass consists predominantly of fat with a minimal blood supply, the risk of bleeding during an endoscopic treatment is small. In case the polyp is rich with vascular structures, endoscopic resection can be troublesome

due to uncontrollable bleeding. Preoperative identifying the place and site of a polyp's stalk, planning of cervical incision opposite to the origin is possible²⁴.

Histological differentiation of FP from IPT is somewhat difficult²⁵. This difficulty is partly due to the lack of exact histological definition of each lesion that gives rise to the nosological confusion²⁶. Key histological findings in establishing the diagnosis of IPT are the co-existence of variable numbers of inflammatory cells and spindle cells, consisting of fibroblasts and myofibroblasts and with varying degrees of fibrosis. This variation in the extent of inflammatory infiltrate and fibrosis suggests that this is a dynamic and evolving inflammatory process²⁷.

The first choice of therapy for this giant, pedunculated, intraluminal masses is surgical excision²⁸. Surgical management is necessary because malignancy can not be excluded preoperatively and this is the only way to get definitive diagnosis and to allow oral feeding. In addition, surgical therapy is recommended because of the progressive nature of the lesion and the underlying risk of asphyxiation and sudden death²⁹.

Management can be complex and varies from endoscopic removal to total esophagectomy, usually is a combination of different types of endoscopic and surgical techniques. Endoscopic removal should be reserved for small, pedunculated tumors without evidence of muscularis propria involvement on EUS. Endoscopic removal has rarely been reported for giant esophageal inflammatory fibrous polyps because the procedure is technically demanding and hemostasis is difficult to ascertain, but it is possible³⁰. Surgical excision is preferable by a left-sided cervical approach and, when tumor is too big, removal should be done through gastrotomy by open access or in laparoscopic way^{9, 31, 32}. In case of a large-size polyp, a thoracotomy may be necessary³³. If there is no stalk, the operative enucleation should be recommended³⁴. Complete surgical resection by esophagectomy, whether partial or total, should be the procedure of choice for large and obstructing esophageal IPTs or any tumor with muscularis propria involvement, decreasing the risk of recurrence³⁵. Nowadays, a minimally invasive approach is more often used in esophageal surgery for these challenging cases³⁶. In our case, we succeed to remove complete tumor without esophagectomy, using a bi-approach.

Local excision of FP is curative and recurrence after resection is very rare. However, there are reports on recurrent FP that recurred within years. Some authors believe that residual tissue around the pedicle's base may cause recurrent polyp formation, which hypothetically can be the reason for recurrence. Local recurrence of giant IPT is rare but may occur if there is incomplete resection of the lesion³⁷. Due to the risk of recurrence, patients should undergo to endoscopic and radiological surveillance for several years³⁸.

In the literature, there are several reports in which corticosteroids were successfully used for the treatment of IPT³⁹. The use of chemotherapy and radiation for IPT treatment is still controversial⁴⁰.

Conclusion

The presented case illustrates the complexities involved in diagnosing and the management of giant esophageal pedunculated tumors. This unique lesion starts in the upper

esophagus and has histopathological picture of FP with clinical manifestations and behavior of IPT, i.e. young ages, fever, malaise, weight loss, and thrombocytosis that resolved after operation. To our knowledge, such a case has not been described in the literature before.

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