The aim of the study was to consider the qualifications necessary for the procedure, the surgical treatment, and the long-term results.

Material and methods. During the period between 1990 and 2005, twenty-five patients with benign esophageal tumors underwent treatment. Clinical symptoms, the size and localization of the tumor, and histological type were considered in cases of patients qualified for surgical intervention. Not all patients with esophageal leiomyomas were subjected to preoperative verification. Intraoperative endoscopic examinations identified the pathology, especially in cases of small lesions.

Results. Patients diagnosed with esophageal leiomyomas were subjected to tumor enucleation. In cases of Abrikosow’s tumor (three patients), the tumor was removed with the esophageal wall. Four patients underwent endoscopic resection. Esophageal fistulas during the perioperative period were not observed. Mortality was not observed during patient hospitalization. None of the cases presented recurrence or malignant transformation.

Conclusions. Surgical treatment is the method of choice for treating benign esophageal tumors. During the postoperative period, neither disease recurrence nor malignant transformation was observed.

Key words: leiomyoma of the esophagus, perioperative cyst, Abrikosow’s tumor, endoscopic excision

Benign esophageal tumors are rarely diagnosed, because they are in most cases asymptomatic. These tumors are often incidentally diagnosed during endoscopic examinations of the upper digestive tract. The most common lesions include leiomyomas, peri-esophageal cysts, and Abrikosow’s tumor. The remaining rarely-described tumors include fibroma, neurofibroma, neurinoma, and vascular and inflammatory tumors.

Microscopic differentiation of tumors distinguishes the following types: epithelial, non-epithelial, and heterotopic. Additionally, tumors can be divided into intramural and extramural lesions (1).

Potentially malignant digestive tract tumors have been defined as gastrointestinal stromal tumors (GISTs), and they arise most commonly in the stomach.

Complete diagnostics of benign tumors should include radiological examinations, upper digestive tract endoscopy with concomitant endosonography, and computer tomography of the thorax. The presence of symptoms and suspicion of malignancy are indications for surgical management. Although malignant transformation is rare, it should not be excluded (2).

MATERIAL AND METHODS

A total of 27 patients, including 16 (59%) males and 11 (41%) females, were treated for benign esophageal tumors at the Department of Thoracic Surgery between 1990 and 2005. Patient age ranged between 17 and 70 years, and the mean patient age was 58.6 years.

Group I was comprised of 16 (59%) patients with leiomyomas (11 male and 5 female patients, aged between 17 and 57 years).

Group II was comprised of four (14%) patients with peri-esophageal cysts (2 male and 2 female patients, aged between 17 and 55 years), whereas group III was comprised of seven (26%) patients with Abrikosow’s tumor (3 male and 4 female patients, aged between 35 and 70 years).
Symptoms

The most common symptoms for patients in group I (16 patients) were pain, burning, and the sensation of the presence of a foreign object in the epigastrium in 11 (69%) patients, pain and burning in the chest in three (19%) patients, and dysphagia in two (12%) patients.

The most common symptoms for patients in group II (4 patients) were abdominal discomfort and the sensation of burning and heaviness in the stomach for two patients and two patients were asymptomatic.

Group III (7 patients) patients complained of nonspecific symptoms, including a burning sensation in the epigastrium, heartburn, and lack of appetite.

Diagnostics

All patients were subjected to the following examinations: chest X-ray, esophageal barium enema, computer tomography of the thorax, and upper digestive tract endoscopy. In addition, one patient underwent esophageal endosonography.

During esophageal endoscopy, histopathological samples were collected from 21 patients. Routine biopsies to obtain preoperative verification were not performed on every leiomyoma patient. However, biopsies were performed in 10 patients, and these patients were qualified for surgical intervention diagnosis no earlier than 14 days after collection of the histopathological sample. The remaining six patients qualified for surgery on the basis of endoscopy and imaging examinations without biopsies. Patients with Abrikosow’s tumor and peri-esophageal cysts underwent biopsies during endoscopy.

Treatment

General anesthesia was initiated after patient intubation and was followed by the introduction of the endoscope into the esophagus for tumor identification. Intraoperative endoscopy was performed in all patients, and it was especially helpful for identifying small tumors (1 cm in diameter). All patients underwent right-sided posterolateral thoracotomies. Leiomyomas and peri-esophageal cysts were subjected to enucleation, and the muscular layer was closed using Vicryl 3/0 sutures. After tumor excision, endoscopy was performed to determine the tightness of the closure. Abrikosow tumors exceeding 1.5 cm required the excision of the esophageal wall. Four patients were subjected to endoscopic removal of the esophageal lesion (1–1.4 cm in diameter). All patients underwent intraoperative verification to confirm the diagnosis, and this verification aided decisions concerning the method of treatment.

RESULTS

The most common symptoms included nonspecific abdominal pain in 20 (74%) patients, dysphagia in two (7.5%), retrosternal pain and heartburn in three (11%), and a lack of symptoms in two (7.5%). Of the 16 patients with leiomyomas, 12 (75%) presented with lesions in the thoracic lower third of the esophagus and four (25%) in the middle part of the esophagus (fig. 1). Endoscopic examination revealed the presence of an isolated, submucosally-located tumor. Twelve (75%) patients had tumors with an oval shape, three (19%) had tumors with a corkscrew shape, and one (6%) had a tumor with a horseshoe shape. The diameter of the lesions ranged between 1 and 10 cm. All patients with leiomyomas were subjected to incision of the muscular layer and tumor enucleation (intramural localization). Except for one lesion localized to the thoracic part of the esophagus, most peri-esophageal cysts and Abrikosow tumors were localized to the lower third of the esophagus. Cysts were 2-3 cm in size (fig. 2), while Abrikosow tumors size ranged between 1 and 2.2 cm (fig. 3). Table 1 presents
the clinical and pathological characteristics of benign esophageal tumors.

No patients showed mucous membrane damage after leiomyoma enucleation. Even for patients undergoing esophageal wall excision, this finding was confirmed by means of intraoperative endoscopy. Esophageal fistulas were also absent. Five of the operated patients developed atelectasis; two required bronchial aspiration, and four presented with pleural effusions. After esophageal wall excision, leakage was not observed in patients with Abrikosow tumors during control X-ray examinations.

Four patients were subjected to endoscopic removal of the esophageal lesion without ensuing complications. Postoperative complications after peri-esophageal cyst removal were also absent. Patient hospitalization ranged from 2 to 12 days (mean-10.5 days).

DISCUSSION

Leiomyomas most commonly localize in the esophagus, and they constitute 10% of all gastrointestinal leiomyomas. Autopsy analyses show that their frequency of occurrence ran-

| Table 1. Clinical and pathological characteristics of treated patients |
|---------------------------------|-----------------|-----------------|
|                                | Leiomysoma       | Peri-esophageal cyst | Abrikosow's tumor |
| Number of pts.                 | 16              | 4               | 7               |
| Endoscopic localization:       |                 |                 |                 |
| a) middle                      | 4               | 1               |                 |
| b) lower part of the esophagus | 12              | 4               | 6               |
| Size of the tumor              |                 |                 |                 |
| 9 – 1.2-1.5 cm                 | 3 – 2 cm        | 3 – 2-2.2 cm    |
| 4 – 1.7-2.2 cm                 | 1 – 3 cm        | 2 – 1.2 cm      |
| 1 – 2.8 cm                     |                 | 1 – 1.4 cm      |
| 1 – 3.5 cm                     |                 | 1 – 1cm         |
| 1 – 10 cm                      |                 |                 |
| Symptoms                       |                 |                 |                 |
| non-specific                   |                 |                 | non-specific    |
| two cases-dysphagia            |                 |                 | non-specific    |
| Treatment:                     |                 |                 |                 |
| a) surgical excision           | enucleation      | enucleation     | a) local excision of the esophageal wall and tumor – 3 patients |
| b) minimally invasive          |                 |                 | b) endoscopic polypectomy – 4 patients |
| Localization in relation to the esophageal wall | intramural | extra-esophageal | submucosal |
| Hospitalization                | 5-10 days       | 3-5 days        | a) 12 days      |
|                                |                 |                 | b) 2 days       |
Benign esophageal tumors

Benign esophageal tumors and cysts are the second most common benign tumor of the esophagus, and they are diagnosed five times less frequently than leiomyomas (5). Abrikosow’s tumor is diagnosed in 8% of patients with esophageal localization; it thus accounts for 2% (6, 7, 8).

Clinical symptoms are connected with unspecific abdominal epigastric pain, esophageal discomfort, sensation of a foreign body, and retrosternal burning and pain. Some authors have reported the absence of symptoms in 50% of patients with diagnosed esophageal leiomyomas (1, 9). Dysphagia was only observed in patients with leiomyomas. Mutrie and co-authors observed that dysphagia developed in patients with tumors exceeding 5.3 cm (9). The diagnosis of patients with peri-esophageal cysts was established 6-12 months after the first visit to the physician. Symptoms were nonspecific.

Because they are asymptomatic, Abrikosow tumors are most often incidentally diagnosed during endoscopic examinations. Some authors mention dysphagia in case of tumors exceeding 1 cm (10).

Esophageal leiomyomas are usually diagnosed in patients between the ages of 20 and 59 years, and they are more frequent in men than women. Lesions are usually isolated, although multiple changes are possible in 10% of cases (11, 12). These lesions derive from the proper muscular layer, but they can occasionally derive from the muscular layer of the mucous membrane. Thus, intramural localization is observed in 97% of cases. Polipoids constitute 1% and extra-esophageal localization is observed in 2% of cases connected with mediastinal structure diseases. Histologically, leiomyomas are derived from smooth fibers with long, fusiform processes and calcifications inside the tumor (1).

Endoscopy reveals that lesions are usually localized submucosally, and rarely do they form as esophageal polipoids. In 80% of cases, leiomyomas are localized to the middle and lower part of the esophagus. The upper part is dominated by skeletal cross striation (13).

Modern diagnostic examinations, such as classical endoscopy, endosonography (EUS), and computer tomography (CT), enable the precise determination of lesion localization, size, and type. Further, these techniques offer the possibility of sample collection for histological verification. Bonavina and co-authors noted that mucous membrane damage occurred more frequently during the surgical procedure after a biopsy esophageal. Thus, preoperative verification should not be performed in every case of esophageal leiomyoma (14, 15). Mutrie and co-authors have also confirmed the above-mentioned findings (9).

Surgery is the therapeutic method of choice. Standard management consists of the enucleation of the leiomyoma, and esophageal resection is rarely performed (16).

All patients were subjected to right-sided postero-lateral thoracotomy. Other authors use the thoraco-abdominal approach, left-sided thoracotomy, and laparotomy (9, 14). After general anesthesia and intubation, the endoscope was introduced into the esophagus to identify the lesion. This process facilitated the surgical procedure, especially in case of patients with small tumors. Surgery consisted of leiomyoma enucleation and closure of the muscular layer. Bardini and co-authors suggested the possibility of pseudodiverticulum occurrence after enucleation during the thoracoscopic procedure. Bonavina and co-authors also reported this finding following classical surgery (17, 14). Intraoperative endoscopy enabled the determination and confirmation of esophageal mucous membrane closure tightness. Other authors also reported its usefulness during leiomyoma enucleation (15, 18). Izumi and co-authors introduced an esophagoscope with a balloon and performed an expulsion of the leiomyoma (18).

Minimally-invasive methods, such as videothoracoscopy (VTS), laparoscopy (LAP), and the da Vinci system first used in 2004, are becoming more common (19). Von Rhaden and co-authors compared classical surgery (the thoracotomy approach) with minimally-invasive methods to demonstrate that both methods are equal; the choice of which to use depends on the experience of the surgeon. The duration of the procedure was similar, but shorter hospitalization and less pronounced symptoms were observed after minimally-invasive procedures. According to the authors, the transhiatal laparoscopic approach can be used in cases of tumors localized to the lower third of the esophagus (20).

Peri-esophageal cysts constitute congenital embryologic residues. Due to the pathological evaluation, one can distinguish duplication cysts or, more rarely, inclusion cysts (1, 21, 15).
They are recognized as extramural tumors, and they are localized to the lower third of the esophagus (15). Possible complications include bleeding, perforation, and dysphagia; thus, their excision is recommended (1). Treatment consists of enucleation and closure of the esophageal wall. During the perioperative period, complications did not develop and recurrence was not observed. Some authors recommend cystic puncture (22). Such treatment is not justified due to the possibility of disease recurrence, threatening complications, and malignancy (23).

Abrikosow’s tumor is derived from Schwann cells; while it is usually benign, metastasis is possible in 1-2% of cases. The tumor rarely accompanies esophageal carcinoma (24, 25). This tumor is most often localized to the lower third of the esophagus (26). Local excision is the therapeutic method of choice, although one occasionally observes excision of the whole esophagus (21, 25, 27). Local recurrence is noted in 0 to 10% of cases (8, 28). Local excision of the esophageal wall with the tumor was performed in three cases. The postoperative period proved uneventful. Recurrence and malignant transformation were not observed. In four cases, the lesions were endoscopically removed using a bi-channel endoscope and loop. The tumor had a 1.2 cm diameter in two patients, a 1 cm diameter in one, and a 1.4 cm diameter in one. Hyun and co-authors described endoscopic resection of benign esophageal tumors in 62 patients. Loop polypectomy was performed in 36 patients, whereas mucosal incision and enucleation were performed in 25. Only one patient was subjected to partial removal of the lesion; this procedure was necessary due to adhesions of the tumor and submucosa. The diameter of the removed lesions ranged between 0.6-7.5 cm (mean – 1.9 cm) (29). Kajiyama and co-authors described nine patients with tumor diameter limited to 2 cm who were subjected to endoscopic resection (30).

Surgical treatment is recommended for tumors with a diameter exceeding 1 cm, whereas lesions <1 cm should undergo endoscopic surveillance via biopsy (9, 27, 31). Voskuil and co-authors recommend endoscopic examinations with biopsies once a year. These authors caution that the examination may prove unreliable because the lesion may be located submucosally. Endosonography may prove decisive due to its diagnostic accuracy of 95% (27). Due to the character of the tumors, diagnostics may prove difficult. Therefore, the above-mentioned tumors should be differentiated from gastrointestinal stromal tumors (GISTs) and squamous cell carcinomas (32).

CONCLUSIONS

1. Benign esophageal tumors are rarely diagnosed, and they should be differentiated from malignant lesions.
2. Surgical treatment is the method of choice for treating these tumors. It consists of local excision of the benign esophageal lesion and produces good therapeutic results without posing difficulties. The choice of the method depends on the tumor type, size, and localization in addition to the therapeutic potential of the surgical department.

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Benign esophageal tumors are rarely diagnosed. Autopsy investigations demonstrate an occurrence of 0.49-0.59% for such cases, and this accounts for 5% of all esophageal tumors (1). Most often, these tumors occur as intramural lesions covered by a normal mucous membrane. Leiomyomas, occurring in 70-80% of cases, are the most common benign esophageal tumors (1). However, stromal tumors, sarcomas, and melanomas should be considered during differential diagnosis. More than 85% of patients with benign esophageal tumors are asymptomatic, and the lesion is often accidentally diagnosed during radiological or endoscopic examinations (1). Endoscopic ultrasonography offers the most valuable examination when the indications and therapeutic method of choice for esophageal intramural tumors are considered. The present study does not describe the characteristics of the endoscopic or ultrasound esophageal lesions. Some features of the tumor, such as a size exceeding 3 cm, follicular shape, ulceration depth >5 mm, heterogenic echogenicity, and presence of an echoless area, may suggest the possibility of malignancy (2). Indications for the excision of the intramural tumor depend on the presence of symptoms and histologically-confirmed pathology that exclude malignancy.

Benign esophageal tumors


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COMMENTARY

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The excision of asymptomatic and small (<3 cm) tumors may raise controversy (2). In selected cases, patient cancerophobia may be considered as a relative indication. Enucleation of the tumor is the therapeutic method of choice. The possibility of endoscopic resection should be considered for cases of tumors localized to the internal side of the muscular layer of the esophagus (3). When the tumor is bound to the muscular layer of the esophagus, enucleation from the thoracic approach is the method of choice. This study lacks clear criteria qualifying patients for either endoscopic or surgical treatment. Due to the risk of perforation, biopsies should be avoided in cases qualifying for surgical enucleation in which esophageal tumors are covered by a normal mucous membrane (2). Clear qualification criteria for performing a biopsy were also absent in the study. It is curious that sample collection was followed by therapeutic procedures without histopathological confirmation. Exposure of the patient to invasive examinations, such as endoscopy with biopsy sampling, in the absence of histopathological confirmation of the lesion’s character can lead to both complications connected with the procedure and erroneous decisions concerning therapy.

Minimally-invasive techniques are being used increasingly more often in esophageal surgery in recent years (2, 4). The enucleation of benign esophageal tumors can be performed by means of thoracoscopy, in which patients are placed on their left side in a prone (2, 4). One should not forget about the indications for esophageal resection, which might prove to be a necessity for large tumors, tumors whose annular shape renders enucleation impossible, numerous intramural tumors, and tumors with a high risk of malignancy.

The present study demonstrates the great experience of the Authors in the diagnosis and treatment of a rarely-occurring group of esophageal diseases. I do not agree with the second conclusion. The choice of the therapeutic method only partially depends on the experience of the surgical center. The treatment of benign neoplastic lesions should depend on tumor type, size, and localization in addition to patient condition.

REFERENCES


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