

Endoscopic resection of an esophageal leiomyoma with overlying dysplasia without specialized equipment

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Abstract

Leiomyomas are rare benign esophageal tumors. Association of this subepithelial lesion with abnormal epithelium is rarer. Endoscopic mucosal resection is an alternative to surgery for removing suitable mucosal and submucosal lesions from the gastrointestinal tract. This procedure is seldom performed in developing countries due to limited equipment and expertise. We describe a case of esophageal leiomyoma with overlying dysplasia in the mid esophagus that was completely removed endoscopically in a developing country without the standard accessory equipment. Traditional thoracotomy would have been associated with higher cost and morbidity.

Key words: Dysplasia, endoscopic resection, esophageal leiomyoma

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Introduction

Esophageal leiomyomas are rare, but are the most common benign esophageal tumors and may be confused for esophageal cancer.^[1] The presence of a normal overlying mucosa is helpful in ruling out cancer.^[2] Traditionally, esophageal leiomyomas are treated by surgical removal via thoracotomy for tumors in the upper two-thirds of the esophagus, but increasingly less invasive methods are now being used with significant reductions in morbidity and hospital stay.^[2] These reductions are essential in patients with co-morbidities. The development and use of these techniques have been largely limited to some developed countries. Endoscopic resection is an alternative to surgery used for removal of early mucosal and submucosal lesions

in the gastrointestinal tract, mainly in the esophagus, stomach, and colon.^[3] This technique has been applied to leiomyomas of the esophagus.^[4] However, apart from endoscopes, it requires specialized accessory equipment that are usually unavailable in resource-poor settings. The relatively late presentation of a majority of patients in such areas also makes most of the lesions seen unsuitable for this form of therapy.

It is against this background that we present a case of a 53-year-old diabetic and hypertensive woman who had an endoscopic resection of an esophageal tumor. In this report, we describe what was done.

Case Report

A 53-year-old woman with the previously diagnosed peptic ulcer disease had repeat endoscopy for epigastric pain. At

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Figure 1: Endoscopic findings: Two subepithelial coalescing lesions with intact overlying mucosa

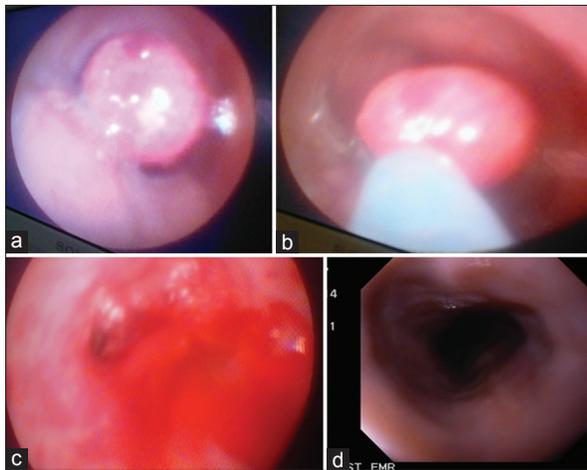


Figure 2: Endoscopic resection procedures and follow-up. (a) After rubber band ligation; (b) lesion captured with a snare; (c) slight bleeding from bed after removal; (d) site of endoscopic mucosal resection 6 months after

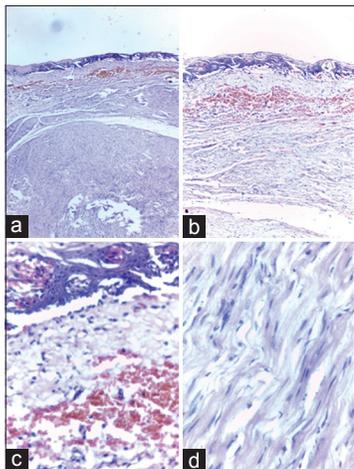


Figure 3: Histology. (a) Esophageal leiomyoma arising from muscularis propria (H and E, ×4); (b) mucosa at increased (H and E, ×10); (c) cellular details of epithelium showing dysplasia (H and E, ×40); (d) cellular details of smooth muscle of leiomyoma (H and E, ×40)

endoscopy, apart from marked inflammation of the antrum and duodenum, two closely related polypoid lesions, noticed in the midesophagus were biopsied. Histological examination revealed dysplasia. She had been diagnosed with hypertension and diabetes mellitus for 10 and 3 years respectively. The gastroduodenitis resolved with triple therapy, but repeat endoscopy almost a year later showed that the esophageal masses had slowly increased in size and were coalescing while mucosal biopsies maintained dysplasia on histology. Three therapeutic approaches were considered based on skills and equipment available, of which the third was chosen: (a) Thoracotomy, (b) video-assisted thoracoscopic surgery (VATS), and (c) endoscopic resection.

The procedure was carried out as follows. The patient had an initial endoscopy with topical lignocaine spray to evaluate the lesions that were 25 cm from the incisor teeth [Figure 1]. Then under conscious sedation, a modified multiple esophageal variceal banding device (Cook Endoscopy 4 Shooter Saeed Multiband Ligators) was introduced with the fiber optic gastroscope (Pentax fiber optic TD-34W) and suction was applied to lesions. Two rubber bands placed on the coalescing lesions resulted in a single polypoid mass. The endoscope was then reintroduced without the banding device, and through the biopsy channel a polypectomy snare (Olympus SnareMaster Disposable Electrosurgical Snare) was introduced and the lesion was snared and removed with some difficulty utilizing diathermy [Figure 2]. The mass removed measured 2.5 cm. Minimal peri-interventional bleeding stopped spontaneously, and there was no perforation. However patient complained of pain on swallowing, and was admitted and placed on nil per mouth. Blood sugar and blood pressure levels measured were within normal limits. By the next day, she commenced clear fluids, and a day after, resumed a normal diet as the pain had resolved. After she had commenced normal diet, she was discharged. Histology of resected specimen was leiomyoma: a nodular round mass of proliferating smooth muscle cell fascicles interlacing with each other, overlying dysplastic epithelium was noted, but did not extend beyond the tumor [Figure 3]. Immunohistochemistry was unavailable. Follow-up endoscopy 6 months after the resection revealed a normal esophagus with no lesions or scarring [Figure 2]. The patient has remained asymptomatic and is still on surveillance.

Discussion

The present case of esophageal leiomyoma with overlying mucosal dysplasia was treated with endoscopic mucosal resection (EMR) without standard specialized equipment. To the best of our knowledge, this is the first reported case of therapeutic endoscopic resection of an esophageal tumor in Nigeria.

Benign tumors of the esophagus are rare but of these rare tumors leiomyomas are the commonest. They are usually located in the mid and distal esophagus and commonly arise from the muscularis propria and less frequently from the muscularis mucosa.^[2,5] About half the patients with esophageal leiomyomas are asymptomatic.^[2] The issue of whether leiomyomas undergo malignant change is controversial, the risk of malignant transformation if ever, is minimal.^[2,5,6]

Dysplasia in the mucosa overlying esophageal leiomyoma is rare. Endoscopic biopsies of the mucosa in our patient revealed dysplasia. Although squamous carcinoma overlying benign tumors like leiomyomas have been reported in several case reports, dysplasia overlying esophageal leiomyoma is even rarer.^[7] Our hypothesis is that the raised mucosa over the benign leiomyoma undergoes chronic irritation predisposing it to dysplasia that can subsequently become a carcinoma. This agrees with the suggestions by other authors that the co-existence of these epithelial lesions with benign subepithelial lesions is as a result of chronic stimulation.^[7,8]

Advances in medical technology like endoscopic ultrasound (EUS) and narrow band imaging (NBI) improve the diagnosis and characterization of subepithelial lesions and epithelial dysplasia respectively. EUS has revolutionized the management of esophageal submucosal lesions by enabling exact localization and origin, tumor margin, echogenic pattern, and accurate size measurement.^[9,10] Without EUS in our facility, we were unable to determine the anatomical plane of the leiomyoma until it had been removed. NBI utilizes optical filters that improve absorbance and scattering of light, enhancing the appearance of vessels and other structures, thus providing a high contrast of tissue surface during endoscopy. Along with magnifying endoscopes and chromoendoscopy, a close to histological tissue description is possible. These have been used to improve the assessment of esophageal lesions.^[11] We did not have any of these to help evaluate the tumor or esophageal mucosa which led to repeat endoscopies and biopsies.

In addition, endoscopic accessories have been developed for EMR utilizing snares with either injection of solutions like saline to raise the lesion up, or specially designed band ligation equipment to ligate the lesion. Another (EMR) method is the “lift and cut” technique where usually a double channel therapeutic endoscope with a snare and forceps is used.^[3,9] These instruments are not readily available in developing countries. The patient’s comorbidity made the decision to attempt an EMR more attractive. Innovatively the multiple variceal ligating devices and a standard snare were used. Concerns exist that the repeat biopsies may increase the likelihood of hemorrhage, spread and make

lesions more difficult to remove completely at EMR. However, biopsies were the only modality available to us for the evaluation of these lesions and immediate follow-up endoscopy as well as a repeat 6 months later did not show these complications in this patient. Endoscopic resections are ideal for small tumors <2 cm, however, larger tumors up to 7.5 cm have been safely removed.^[12]

VATS had also emerged as an option for the treatment of leiomyoma of the middle third of the esophagus.^[2,13] Absence of necessary equipment and expertise prevented its consideration in this case. The morbidity associated with thoracotomy made it an unattractive choice for this patient.

Conclusion

EMR can be performed successfully in developing countries for suitable esophageal lesions with limited equipment.

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Conflicts of interest

There are no conflicts of interest.

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