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Case Report

A Case of Esophageal Squamous Papilloma: An Unusual Cause of Dysphagia and Hematemesis in a Patient with Concurrent Malignancies

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Abstract

Introduction: The esophageal squamous papilloma (ESP) is a rare cause of dysphagia and hematemesis. The malignant potential of this lesion is uncertain; however, the malignant transformation and concurrent malignancies have been reported in the literature. **Case Description:** We report a case of esophageal squamous papilloma in a 43 years old female who had a background diagnosis of metastatic breast cancer and liposarcoma of the left knee. She presented with dysphagia. Upper gastrointestinal (GI) endoscopy showed a polypoid growth, and its biopsy confirmed the diagnosis. Meanwhile, she presented again with hematemesis. A repeat endoscopy showed that the previously seen lesion had likely broken off, leaving behind a residual stalk. This was snared and removed. The patient remained asymptomatic, and a follow-up upper GI endoscopy at six months did not show any recurrence. **Practical Implications:** To the best of our knowledge, this is the first case of ESP in a patient with two concurrent malignancies. Moreover, the diagnosis of ESP should also be considered when presenting with dysphagia or hematemesis.

Key words: Dysphagia, esophageal squamous papilloma, hematemesis, pre-malignant

Introduction

Esophageal squamous papilloma (ESP) is a rare entity.[1,2] Around 200 cases have been reported in the literature. The usual age of presentation is between 43 and 50 years, with variable male to female ratio.[1,2] ESPs commonly appear as solitary sessile nodules located in the lower or middle part of the esophagus. The size ranges from 2 to 6 mm.[1,2] A few cases with multiple lesions,

called esophageal papillomatosis, have also been reported. Histologically, it appears as a finger-like projection with stratified squamous epithelium and preserved normal cellular or acellular atypia. There are only a few reports of malignant transformation in solitary papillomas. However, exceedingly rare papillomatosis carries significant malignant potential.[2-5] The lesions are generally amenable to endoscopic resection. Lesions smaller than 1 cm can usually be removed with biopsy forceps, but larger lesions require endoscopic mucosal resection.[1,2,4] Recurrence is infrequent after resection.[1,2]

Case Description

A 43-year-old female was referred to the gastroenterology clinic at Shaukat Khanum Memorial Cancer Hospital and Research Centre to evaluate progressive dysphagia to solids for two months, associated with occasional vomiting without blood. She had lost 4 kg weight. Her symptoms of gastrointestinal reflux disease (GERD) were not responding to high dose omeprazole and domperidone. Her past medical history was significant for carcinoma of the left breast with metastatic disease to the liver and uterus. She was treated with chemotherapy and local radiotherapy to the breast. Recently, she was also diagnosed with liposarcoma of the left knee, for which chemotherapy was planned. There was no history of NSAIDs or steroids use. A detailed history was not suggestive of any habits (alcohol or smoking) or any gastrointestinal (GI) malignancies in the family.

Diagnosis and management

The patient underwent esophageal-gastro-duodenoscopy (EGD), which revealed a polypoid growth of around 1.5 cm, at 27 cm from the incisors, which appeared to be twisted upon itself [Figure 1]. Biopsies were taken, which revealed squamous papilloma. Follow-up was planned; meanwhile, the patient presented to the emergency department with multiple episodes of hematemesis. A repeat EGD showed that the previously seen lesion had likely broken off, leaving behind a residual stalk. This was snared and removed [Figure 2]. The biopsy of the stalk confirmed squamous papilloma with spongiosis and chronic inflammation. No dysplasia or malignancy was seen. On six months follow-up, the patient remained asymptomatic. A follow-up EGD revealed only a scar at the site of the lesion.

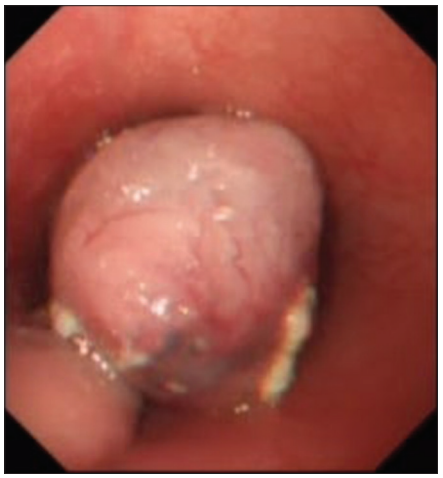

Figure 1: Endoscopic appearance of esophageal papilloma on first endoscopy

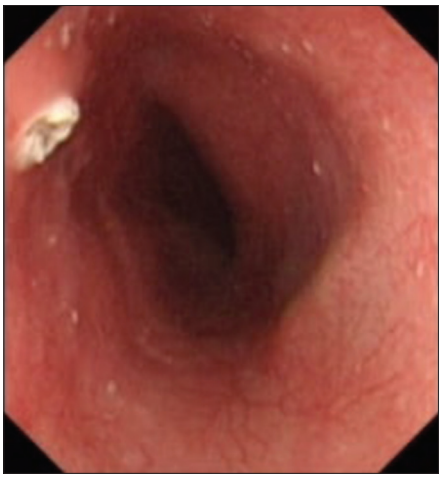

Figure 2: Endoscopic appearance after removal of residual stalk of spontaneously detached esophageal papilloma on subsequent endoscopy

Discussion

ESP is extremely rare, with a prevalence of 0.01–0.23%.[1,2] These are usually solitary lesions in the middle or lower part of the esophagus. The papillomatosis (multiple lesions) is more

exceptional, and around 13 cases have been reported in the literature. In general, patients are asymptomatic, and the lesion is diagnosed incidentally. When symptomatic, the most typical presentation is of GERD, which responds poorly to medical therapy. Dysphagia has been a rare presentation as lesions are too small to cause obstruction (2–6 mm). In a study by Jideh *et al*., only 1 out of 16 patients with ESPs had dysphagia.[1]

Similarly, only a few patients may present with hematemesis.[1,2] Our patient presented with these unusual symptoms of progressive dysphagia and hematemesis, in addition to refractory GERD. This unusual presentation was likely due to the larger size (1.5 cm) of the lesion in our case as opposed to what is reported in the literature (2–6 mm).[1,2] The exact etiology is still unknown, but factors resulting in mucosal injury have been proposed to be responsible in literature. These include chemical irritants, gastro-esophageal reflux disease, and human papillomaviruses (HPV).[1,2] Our patient had refractory GERD, and the biopsy also showed chronic inflammation. HPV status was not checked in our case. The role of HPV infection is controversial.[1,2] A study by Panthem *et al*. reported its association with ESPs in 47.4%. In contrast, according to another study, the association was not statistically significant.[6-8]

The malignant potential of these lesions is uncertain. There are only a few reports of malignant transformation in solitary papillomas. However, exceedingly rare papillomatosis carries significant malignant potential.[2-6] In a French study, 1 out of 78 cases (1.3%) of esophageal papilloma developed into squamous cell carcinoma at a follow-up of 2 years.[2] In our patient, the biopsy of the lesion was negative for dysplasia or malignancy. However, the patient had concurrent cancer at other sites (metastatic breast cancer and liposarcoma of the knee). Various treatment modalities, including endoscopic resection with snare polypectomy, biopsy forceps, and cautery, have been used for single small lesions. The patients with extensive ESPs have been treated with endoscopic resection, radiofrequency ablation, cryotherapy, and laser, as well as oesophagectomy in a few cases.[1-6,9,10] The clinical course in our patient was unusual as the lesion was shed off spontaneously, leaving behind a residual stalk, which was snared and removed endoscopically.

In conclusion, we suggest that the diagnosis of squamous papilloma should be considered in middle-aged patients who present with refractory GERD, non-progressive dysphagia, or hematemesis. There are no particular guidelines for treatment or surveillance; however, a complete removal is suggested considering the reports of malignant transformation.

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Author contributions

Conceived and designed the analysis: MS, MZS, SM, MAY, Collected the data: MS, MZS, JI, Contributed data or analysis tools: MS, MZS, JI, SM, Performed the analysis: MS, Wrote the paper: MS, MZS, MAY.