

## Total Esophagectomy and Endoscopic Radiofrequency Ablation for a Case of Diffuse Esophageal Papillomatosis: Case Report

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### Abstract

Esophageal squamous papilloma (ESP) is a benign epithelial lesion. ESP is extremely rare and it has been described in few cases in literature. The etiology of ESP may be associated to chronic mucosal irritation and HPV infection. It is usually asymptomatic and mostly discovered as an incidental finding during upper gastrointestinal endoscopy. It is considered a benign neoplasia but recent reports have stressed the potential malignant evolution of these lesions. A 53 year old caucasian woman presented with a four years history of dysphagia not responsive to medical therapy. At upper endoscopy, esophageal mucosa appeared hyperemic and completely covered by numerous (>100) sessile pedunculate papules. Biopsies demonstrating atypical epithelial proliferation confirmed the diagnosis of esophageal squamous papillomatosis with low grade and focal high grade dysplasia; *in situ* hybridization studies for HPV were performed and it was positive for the type 16. We report a case of diffuse esophageal papillomatosis with histologic and microbiologic findings of genotype 16 HPV and Lichen Planus infection successfully treated with an Ivor Lewis Esophagectomy and a second look endoscopy by radiofrequency ablation.

**Keywords:** Total esophagectomy; Endoscopic radiofrequency ablation; Diffuse esophageal papillomatosis

### Introduction

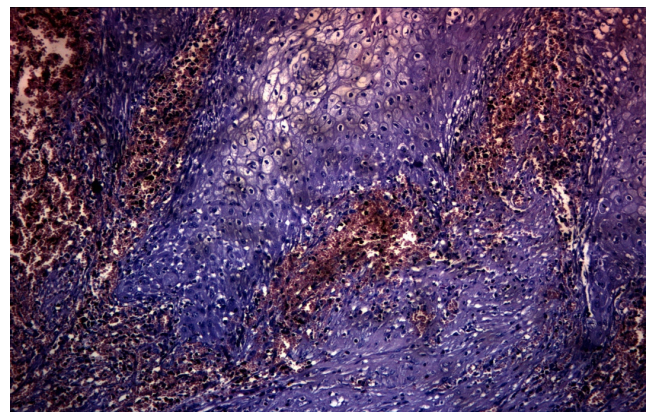
Although first described [1], squamous papilloma of the esophagus is a rare endoscopic finding, occurring in 0.01% of individuals at autopsy and 0.07% of patients during routine endoscopy [2]. Esophageal papillomatosis is a very rare condition and it has a benign clinical course. The natural history of papillomatosis is controversial, with early reports suggesting that it was a benign condition and that endoscopic surveillance was unnecessary [2]. More recently, there have been reports of malignancy in cases of esophageal papillomas. This has led some authors to recommend that esophageal papillomatosis be considered a premalignant condition with the potential for the development of squamous cell carcinoma [3]. Some studies have pointed to the possible presence of malignant tumors in association with esophageal papillomatosis.

Among the viral infections that have proven to play an active role in esophageal carcinogenesis, persistent infection with human papilloma virus (HPV) is thought to have the greatest importance but clear association has not yet been proven. Human papilloma virus (HPV) is an oncogenic, double-stranded, DNA-virus that infects skin and mucosa. Usually associated with benign papillomas, mucosal HPV Infection is one high-risk types, such as HPV 16 and 18 can rarely progress to dysplasia and cancer, a process that takes a long time to occur [4,5].

We describe a case of diffuse esophageal papillomatosis with histologic and microbiologic findings of genotype 16 HPV and Lichen Planus infection successfully treated with an Ivor Lewis Esophagectomy and a second look endoscopy by radiofrequency ablation of the new lesions.

### Case report

A 53 year old Caucasian woman was evaluated in Hospital for weight loss and progressive solid food dysphagia not responsive to proton pump inhibitors. Personal Health revealed genital and oral Lichen Planus and for HCV infection. Two years before, the first gastroscopy was negative.



**Figure 1:** Biopsy of the esophageal lesions. Acute inflammation associated with nuclear changes (X100 magnification).

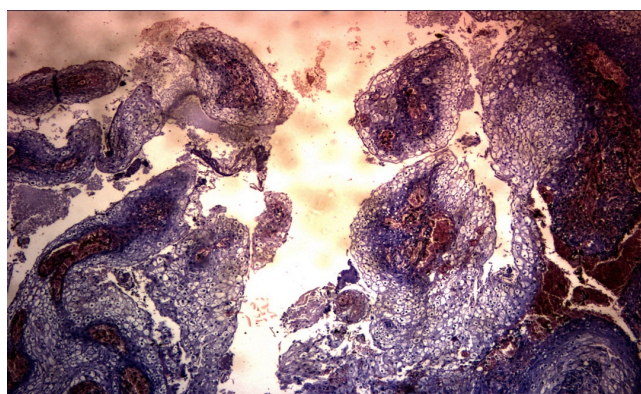
A second endoscopy, performed for solid food impaction, revealed extensive esophageal nodularities and hyperemic, spontaneously bleeding mucosa and it demonstrate multiple flesh coloured pedunculated papules, involving the entire circumference.

Focal high grade dysplasia and intraepithelial inflammation associated with nuclear changes were found by biopsies; due the possibility of human papilloma virus (HPV) infection, *in situ* hybridization studies for HPV were performed and it was positive for subtype 16.

The patient underwent endoscopic ultrasound that demonstrated that the changes were limited to the mucosa without penetration into the muscularis mucosa.

A chest computed tomography showed dilated appearing esophagus with a thickening of the mid-distal wall, extending from 15.5 cm to the gastroesophageal junction. Due to the severity of clinical findings we have made a total esophagectomy with gastro-pharyngeal anastomosis.

On histological examination of the esophagus, an extensive papillomatosis with diffuse low grade and focal high grade dysplasia was found. The inflammatory infiltrate was chiefly lymphocytic and it formed a dense band, typically observed in Lichen Planus (LP) (Figures 1 and 2). Thus we diagnosed diffuse papillomatosis of the esophagus complicated by esophageal lichen planus and HPV 16 infection in patient HCV positive.



**Figure 2:** Histological findings of the lower esophagus demonstrating papillomatous squamous proliferation with keratosis (X50 magnification).

Initial follow-up, six months after esophagectomy, was negative for new papillomas but positive only for granulation tissue. Follow-up endoscopy, one year later, revealed three flesh-coloured pedunculated papules at the gastro-pharyngeal anastomosis; we elected to proceed with endoscopic ablative therapy using the radiofrequency.

To radiological follow-up, performed one year after surgery, it has been documented the correct transit of the contrast medium through the stomach pulled up.

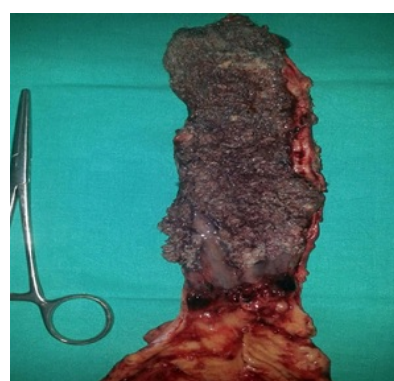
## Discussion

In literature, small isolated lesions have been successfully treated with endoscopic resection, snare polypectomy and cautery [6] but the management of multiple esophageal lesions is more difficult and, due to scarcity reported cases treated in literature, the optimal clinical management of diffuse papillomatosis remains unclear. Photodynamic therapy and radiofrequency ablation may also be useful treatment options [7].



**Figure 3:** Excised segment of the esophagus and stomach.

Surgical resection has even been advocated when malignancy is suspected but not demonstrated in biopsies [8].



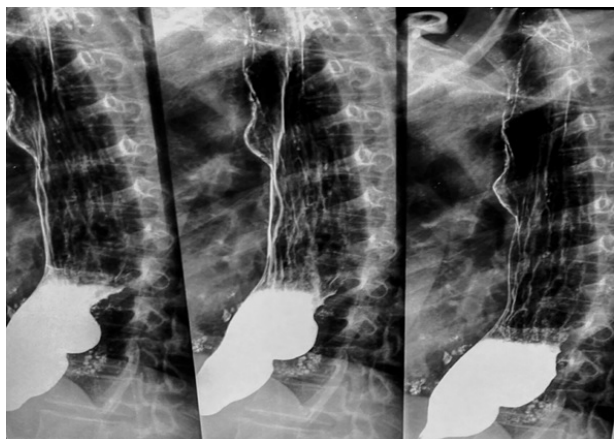
**Figure 4:** Esophagectomy specimen revealed numerous (>100) sessile polypoid mucosal nodularities, to extending from the piriform sinus to the esophago-gastric junction. Their size ranged from 1 to 10 mm.

To our knowledge, the present report describes the first successful removal of multiple esophageal squamous papillomas using total esophagectomy and ablation by radiofrequency in second time of residual lesions (Figure 3). In some case, squamous papillomas have the potential for malignant transformation but the optimal management of these lesions has not been established; however, removal of these lesions during endoscopy using radiofrequency is a reasonable option.

## Conclusion

Our patient, differs from most cases of esophageal papillomatosis reported in the literature in that, endoscopy therapy using injection therapy of an antiviral drug or removal by piecemeal polypectomy techniques used only in cases with a limited number of lesions, instead, in this case report the massive amount of papillomatous tissue

occluding the esophageal lumen and it required treatment with surgery.



**Figure 5:** Radiological follow-up, performed one year after surgery, it has been documented the correct transit of the contrast through the stomach pulled up.

In the majority of the cases, squamous cell papilloma (SCP) of the esophagus can be removed using endoscopic biopsy forceps because most are only a few millimeters in size (Figure 4). Larger papillomas

can be removed using endoscopic mucosectomy (Figure 5). This case underscores the importance of optimizing treatment results by combining the use of surgery and the endoscopic modalities for the ablation of residual lesions.

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