Endoscopic mucosal resection of esophageal squamous papillomatosis

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A 62-year-old woman was referred for investigation of heartburn. She did not have any other significant medical history and denied any other symptoms including weight loss, dysphagia, odynophagia, regurgitation and hematemesis. Her only medication was pantoprazole 40 mg daily for her gastroesophageal reflux disease. Endoscopy was performed with a GIF-Q180 gastroscope (Olympus Optical, Japan), which demonstrated multiple flesh-coloured pedunculated papules at the gastroesophageal junction (Figure 1). Biopsies demonstrating atypical epithelial proliferation confirmed the diagnosis of esophageal squamous papillomatosis, without the presence of dysplasia or carcinoma. A polymerase chain reaction study was negative for herpes simplex virus. Following discussion with the patient, an extensive endoscopic mucosal resection (EMR) incorporating all of the lesions was performed using a band mucosectomy device (Duette, Cook Medical, Ireland) (Figures 2 and 3). Histology from the EMR specimens was similar to the original mucosal biopsy results. No immediate or delayed complications occurred.

DISCUSSION

Although first described in 1959 (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). There is a reported variability in the geographical distribution of this lesion, with a high prevalence in Europe, accounting for approximately 75% of the reported cases. This lesion is usually an incidental finding as a small (<0.5 cm) single lesion, most commonly located in the distal esophagus (3).

Squamous papillomas are typically reported as fleshy-pink in colour, with a soft to warty texture and exist in either sessile or pedunculated form. While the typical endoscopic appearance is a single, round sessile lesion, they can present with multiple esophageal lesions, with rare reports of papillomas exceeding 1 cm in diameter (4). Histologically, they are characterized by vascular connective tissue stalks branching out from the lamina propria with an overlying hyperplastic squamous epithelium. Normal cellular orientation and differentiation are preserved and, unlike squamous cell carcinoma, atypical mitoses or dysplastic cellular invasion into the submucosa are absent. Squamous papillomas of the esophagus are typically asymptomatic, but can be associated with dysphagia, with clinical improvement after removal (5). The predilection of squamous papillomas for the distal esophagus, in addition to its common association with surrounding changes of histological changes of reflux esophagitis, has supported the theory that chemical irritation from reflux of acidic gastric contents has a role in their development. Chemical induction of esophageal papillomas has been demonstrated in experimental animal models. Chemical agents produce leukoplakia, resulting in verrucous hyperplasia of the epithelium that can lead to papilloma formation (6). Our patient had multiple distal esophageal squamous papillomas, otherwise known as squamous papillomatosis, which is even less common than single papillomas. A literature review identified only nine such cases of squamous papillomatosis, some which were positive for human papillomavirus (HPV). Syrjanen et al (7) demonstrated an association with HPV. However, squamous papillomatosis can be found in the absence of HPV.

Figure 1) Squamous papillomatosis

Figure 2) Endoscopic mucosal resection

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Received for publication June 25, 2012. Accepted July 12, 2012

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 multiple reports of malignancy found within squamous 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 (8). Interestingly, the reported cases of carcinoma involved extensive papillomatosis rather than isolated lesions.

The management of esophageal papillomatosis has not been clearly defined in the literature. Small isolated lesions have been successfully treated with endoscopic resection using biopsy forceps, snare polypectomy and cautery (4). The management of multiple esophageal lesions can be more challenging and, due to the paucity of reported cases, the optimal clinical management of extensive papillomatosis remains unclear. Photodynamic therapy and radiofrequency ablation may also be useful treatment options (9). Surgical resection has even been advocated when malignancy is suspected but not demonstrated in biopsies (10). If no specific treatment is undertaken, surveillance endoscopy should be considered given the potential for development of malignancy. However, the specific surveillance strategy has not yet been characterized. To our knowledge, the present report describes the first successful removal of multiple esophageal squamous papillomas using EMR (Duette band mucosectomy device).

Squamous papillomas of the esophagus are a rare finding during endoscopy, but have the potential for malignant transformation in some cases. Optimal management of these lesions has not been established; however, removal of these lesions during endoscopy using EMR is a reasonable option.

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Figure 3) Post-resection

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