Esophageal papillomatosis complicated by squamous cell carcinoma

A 64-year-old woman was referred for investigation of a thickened esophagus on computed tomography (CT) scanning. She had no upper gastrointestinal symptoms, in particular, any dysphagia or weight loss. Nine years previously she had undergone a bone marrow transplant for myelodysplasia for which she was taking 5-mg prednisolone. Endoscopy with a GIF-Q180 gastroscope (Olympus, Tokyo, Japan) demonstrated a circumferential area of multiple confluent papules extending for 5 cm in the mid-esophagus, with a 2-cm nodule at its distal border. The remainder of the esophagus was normal (Fig. 1, Video 1). Histological examination revealed atypical squamous epithelial proliferation, consistent with papillomatosis, and evidence of invasive squamous cell carcinoma in the nodule (Fig. 2, Video 1). Studies to identify human papillomavirus (HPV) as potential etiology were not performed. The patient has been referred for chemoradiotherapy.

Esophageal papillomatosis is a rare endoscopic finding with an unclear etiology. It has been associated with both HPV and reflux disease [1]. The etiology in our patient may be related to HPV infection secondary to long-term immunosuppression. The typical endoscopic appearance is a single, round, sessile lesion. However, our case, similar to other reports, demonstrates that it might present with multiple lesions [2,3]. Early reports suggested that this was a benign condition and endoscopic surveillance was unnecessary [4]. However as our case demonstrates, confirming other reports, esophageal papillomatosis should be considered a premalignant condition with the potential for squamous cell carcinoma development [2,3]. Interestingly, the reported cases of carcinoma have involved extensive rather than isolated lesions. Small isolated lesions have been successfully treated with endoscopic resection [5]. Due the paucity of reported cases, the best clinical management of extensive papillomatosis remains unclear. If no specific treatment is undertaken, surveillance endoscopy should be considered, given the potential for malignant development. The exact surveillance strategy, however, is not known.

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Fig. 1 Endoscopic images of the esophagus in a 64-year-old woman referred for a thickened esophagus on computed tomography (CT) scan. a Multiple confluent papules with circumferential involvement of mid-esophagus. b A discrete nodule at the distal border.

Fig. 2 a Histological section from a biopsy specimen from one of the papules demonstrating atypical squamous epithelial proliferation consistent with papillomatosis. b Histological section from the distal nodule demonstrating invasive squamous cell carcinoma.

Video 1

Gastroscopy demonstrated a mid-esophageal circumferential area of multiple confluent papules and a 2-cm nodule at the distal border of the confluent area. Histological examination revealed atypical squamous epithelial proliferation consistent with papillomatosis and invasive squamous cell carcinoma in the nodule.
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