Benign oesophageal papillomatosis

A case report with a review of the literature

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SUMMARY An 18-year-old boy presented with a four-year history of dysphagia which had been treated repeatedly by endoscopic removal of papillomata of the oesophagus. Eventually, due to increasing dysphagia and repeated recurrences of the papillomata, resection of the affected lower third of the oesophagus was deemed necessary. The resected segment of the oesophagus was carpeted with numerous benign squamous papillomata. The clinical features, radiographic appearances, and pathology of this extremely rare tumour are presented. Only two acceptable cases of oesophageal papillomata can be found in the literature, making this the third case, and apparently the only one with multiple lesions.

Benign oesophageal papillomata are extremely rare and there is at least one statement that true papillomata of the oesophagus are not seen in humans (Stout and Lattes, 1957). This report presents a case of benign oesophageal papillomatosis and discusses the relevant literature.

Case report

This boy first presented to his family physician at the age of 14 years with a history of increasing dysphagia for solids. At endoscopy numerous warty growths were seen at the lower end of the oesophagus. Endoscopic removal of the largest of these lesions relieved the dysphagia, and the tissue was said to show the presence of a 'benign papilloma' on histological examination.

Over the next four years several admissions to hospital for endoscopic removal of the papillomata were required to relieve his recurring dysphagia. He was admitted to the McMaster University Medical Centre in 1975 on two occasions. On the first of these occasions an endoscopic biopsy showed the presence of a benign oesophageal papilloma. On the second occasion, which followed the first after an interval of three months, he complained of a considerable increase in the severity of his dysphagia, and a detailed assessment was undertaken.

On examination there were no physical signs of

note, except that he was lean but not malnourished (height 171 cm, weight 53 kg). In particular there were no dystrophic changes in the nails, skin or mucosa, and no enlarged lymph nodes could be palpated except in the inguinal region.

Cineradiological studies showed poor distensibility of the lower third of the oesophagus, and an air contrast-oesophagogram revealed a cobblestone pattern of multiple small filling defects in the affected segment (Fig. 1). There was no evidence of a hiatus hernia nor any evidence of reflux. Manometric studies were unsuccessful because the extreme rigidity of the lower oesophagus prevented the passage of the tubes through it. Endoscopy showed the presence of multiple warty papillomata without evidence of ulceration. These findings were considered to be compatible with a diagnosis of benign oesophageal papillomata, but the history of recurrent dysphagia, which had increased in severity, and the poor distensibility raised the possibility of malignant change. It was decided therefore to remove the affected segment.

Using midline epigastric and right thoracotomy incisions, the thickened lower third of the oesophagus was resected with a cuff of stomach at the distal end. Oesophagogastric anastomosis was then achieved.

PATHOLOGICAL FINDINGS

The excised oesophagogastric specimen measured 7 × 4 cm and was of an average 2 cm in thickness.

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Fig. 1 Air-contrast oesophagogram showing the cobblestone effect produced by multiple papillomatous filling defects.

The oesophageal surface was covered with multiple, small, friable, papillomatous excrescences extending over an area 3 cm in width (Fig. 2). Histologically the lesions were made up of multiple papillary projections of the mucosa (Fig. 3). Each papilloma contained a small, delicate central core of fibrovascular tissue and was covered by a thick layer of hyperplastic squamous cells (Fig. 4). Polarity of the squamous mucosa was well preserved, apart from the presence of scattered vacuolated cells near the apices of the papillomata. No atypical nuclear forms were noted and no viral inclusions were seen. A moderate scattered inflammatory infiltrate was present in the lamina propria. The subepithelial muscle was greatly thickened. There was no evidence of invasion. Apart from a small area near the earlier biopsy site there was no evidence of ulceration and no ectopic gastric epithelium was present.

A diagnosis of multiple, benign, squamous papillomata of the oesophagus was made.

FOLLOW-UP

The patient was followed on a regular basis. While he has not suffered from dysphagia he has had symptoms of reflux with ulceration at the anastomosis. No recurrences of the papillomata have been seen.

Discussion

It is clear that the literature on benign epithelial lesions of the oesophagus is confused and many lesions are incorrectly diagnosed as papillomata. It is precisely this confusion that led Totten *et al.* (1953) to state that 'No instance of true papillomata, that is a warty epithelial growth, was encountered in human beings'. This scepticism is reinforced by Stout and Lattes (1957). The only lesions that were acceptable to these workers were the coral-like sessile lesions, with a central core of fibrovascular tissues capped by squamous epithelium, that were seen in animals. Such a definition would exclude inflammatory polypi and reactive changes on the edge of other oesophageal lesions.

Adopting Stout's criteria (Stout and Lattes, 1957; Totten *et al.*, 1953), we believe that the cases of papillomata described by Patterson (1927), Kernan (1927), Ginsburg (1931), McKinney (1931), and Hunt (1937) should, reluctantly, be rejected since their description of the biopsy specimens obtained through the oesophagoscope are relatively inadequate to satisfy the definition provided. Plachta (1962) found two cases of 'papilloma' in a review of 90 benign tumours of the oesophagus but described them as arising from the submucosa and did not illustrate the features of the lesions. Similarly, the review of Moersch and Harrington (1944) provided no details of the lesions seen in three of their patients who were thought to have papillomata. On the other hand, Adler *et al.* (1959) described a solitary oesophageal papilloma in a 73-year-old man that satisfied all the suggested criteria. They also described four additional cases of lesions initially diagnosed as 'papillomata' that were shown on fuller examination to be areas of hyperplasia on the edge of infiltrating tumours of the oesophagus. Weitzner and Hentel (1968) described a particularly convincing case of a solitary lesion in a 70-year-old man discovered at necropsy.

We believe that our case is the third instance of benign oesophageal papilloma that satisfies the suggested criteria and the only case with multiple papillomata (Table). It is notable that, despite the initial fear of malignant supervention, there was no

Fig. 2 Excised segment of oesophagus and stomach showing the pebbled effect produced by the multiple papillomata in the lower oesophagus. (approx. $\times 3.5$)

Fig. 3 Whole mount specimen of cross section of the oesophagus showing the numerous coral-like sessile papillomata. There is very obvious hypertrophy of the muscle, especially in the subepithelial zones to the right of the illustration. Haematoxylin and eosin $\times 10$

Fig. 4 Higher power view of one of the papillomata showing the cap of hyperplastic squamous epithelium and the fibrovascular core. (H and E $\times 170$)

evidence of malignancy on histological grounds in any of the three cases, and that there was no local recurrence after adequate resection of the affected segment of oesophagus (Adler *et al.*, 1959; present case). Minielly and co-workers (1967) described five patients with verrucous squamous-cell carcinoma of the oesophagus. The lesion seen in our patient can

be distinguished from this variant of epidermoid carcinoma by the absence of exuberant papillary fronds, swollen rete pegs, clefting of epithelium, nuclear atypia, and invasion of the subepithelial tissues. It is obvious, however, that an inadequate biopsy of a verrucous carcinoma could be confused very easily with a benign papilloma or *vice versa*.

Multiple oesophageal papillomata are common in swine and cattle and they may be associated with similar papillomata in the oropharynx (Jubb and Kennedy, 1970). In these animals the lesions are thought to be induced by a virus and related to the transmissible genital papillomata which they closely resemble. Despite the lack of demonstrable inclusion bodies in the lesions observed in our patient, we cannot exclude a viral aetiology since no virus cultures were attempted. Persistent papillomatous lesions are known to occur in humans in the perianal region, genitalia, and larynx and are known to have a viral aetiology.

In the process of recovery after severe reflux oesophagitis islands of squamous epithelium that have escaped destruction proliferate and reepithelialise the oesophagus. Occasionally these islets may become hyperplastic—the oesophageal warts referred to by Peters (1966)—and very rarely they may progress to squamous carcinoma. We do not believe that this sequence of events applies in our patient, since there was no history compatible with reflux oesophagitis nor any cineradiological evidence of reflux before resection of the affected segment of oesophagus.

It is difficult to assess the malignant potential of oesophageal squamous papillomata since not only are solitary lesions rare, but multiple lesions are rarer still. Our own experience suggests that the risk is probably small. However, the likelihood of local recurrences and the development of increasing rigidity of the affected segment may make surgical resection the only effective procedure in the long term.

Table Summary of main features in reported cases of oesophageal papillomatosis

Author	Age at presentation (years)	Sex	Site of involvement	Treatment	Follow-up
Adler <i>et al.</i> (1959)	73	M	Lower third, apparently solitary lesion	Surgical resection of lower segment	Survived and symptom free
Weitzner and Hentel (1968)	70	M	Middle third of oesophagus; solitary lesion	Incidental finding at necropsy	—
This case	14	M	Lower third, multiple lesions	(a) Multiple cropping (4 yr) (b) Resection of lower third of oesophagus at 18 yr	Survived; no local recurrence; no dysphagia; ulceration at anastomosis site (2 yr postoperatively)

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