Case study

Esophageal papilloma: A case report of 4 patients

Abstract :

Papillomas are rare benign lesions that can be found in the oesophagus. Being rarely seen is thought to be the reason for the lack of consensus on their management.Through these different cases, we would like to show the clinical and endoscopic difference that papillomas in the oesophagus can present with; and suggest a protocol that will allow better management in the presence of risk of malignant transformation.

KEY WORDS:oesophageal papilloma, upperendoscopy, endoscopic resection

Introduction:

Papilloma of the oesophagus was first described by Adler et al. in 1959 [1]. It is a rare benign tumour of the oesophagus, mostly discovered incidentally at endoscopy, but can also be symptomatic. Its natural history remains poorly understood, and the risk of malignancy exists [2].There is no consensus on treatment or monitoring methods. We report four cases in our department.

Case reports:

Case report 1:

A 54 year old male patient known to be diabetic, on insulin, with a history of decompensated viral B and delta cirrhosis (MELD score of 17 and CHILD- PUGH C 11) admitted to hospital for acute epigastric painendoscopic which did not respond to symptomatic treatment. The upper endoscopy revealed a duodenalulcer with grade II oesophageal varices and hypertensive gastropathy in addition to two small lesions of the middle third of the oesophagus, one was

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pedunculated which was totally resected by cold forceps. The histological analysis showed papilloma.

Case report 2:

A 66 year old male patient, treated for prostatic adenocarcinoma, operated for subependymoma of the 4th ventricle complicated postoperatively by pharyngo-laryngeal paralysis leading to a tracheotomy, and subsequently swallowing problems needing adefinitive gastrostomy. During endoscopic follow-up 10 years after placement of the gastrostomy tube, several centimetric lesions in the middle oesophagus were found. The histo-pathology confirmed the papillomatous nature of the lesions. He subsequently underwent several sessions of argon plasma electrocoagulation of these lesions, which resulted in their almost complete disappearance.

Case report 3:

A 65-year-old female smoker patient known to be hypertensive with a family history of adenomatous polyps underwent an upper endoscopy which revealed an 8 mm pedunculated papilloma in the upper third of the oesophagus. It was completely resected by cold forceps.

Case report 4:

A 64 year old male patient, treated for alcoholic compensated cirrhosis. An upper endoscopy was performed searching for esophageal varices. Surprisingly, 3 small lesions measuring 2-3 mm in the lower third of the oesophagus were found. A biopsy was performed to confirm the diagnosis by cold forceps.

Discussion:

Oesophageal papilloma is a rare epithelial tumour with a prevalence in patients undergoing upper GI endoscopy of 0.01%-0.45%. In the general population, their prevalence is estimated at 0.006%-0.04% based on autopsy series (3-6). The age of onset of papilloma is often around 50 years (7); in our series, the average age at diagnosis of papilloma was 59 years.Several publications, such as TSAI et al (7) and Szanto et al (4), show that oesophagealpapillomas are more frequent in women, whereas other publications, such as Takeshita et al (8) and Huart et al (2), report a higher number in men, as the case in our series.

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There is no much known about the pathophysiology. Two main causes have been discussed and appear to be synergistic [2]: chronic irritation of the oesophageal mucosa due to chemical or mechanical factors, and HPV infection. The mechanism of chronic inflammation can be found in gastro-oesophageal reflux, barrett's esophagus, peptic oesophagitis, and mechanical esophageal trauma such as variceal sclerosis or self-expanding metal prostheses [2]. This coincides with our case reports, most of which present these favourable factors described above.Human papillomavirus (HPV) has also been implicated in the formation of oesophageal papilloma. HPVs are known to cause benign lesions, but can lead to the development of cancer. Infection with human papilloma virus (HPV) has been suggested as a risk factor for oesophageal carcinoma. Some authors suggest a role for HPV in oesophageal carcinogenesis in areas of high incidence of oesophageal squamous cell carcinoma, a role not found in areas of low incidence. The HPV subtypes involved appear to be high-risk HPVs such as HPV-16 and 18 [2]. Some studies, shown that the esophageal papilloma, has no relation with HPV.Its role is still, very much debated [3]. The prevalence of HPV detection in oesophageal papilloma varies between 0% and 87.5% in the various series published [8]. Takeshita et al found a prevalence of 10.5% of HPV positive in his series, while Bohn et al reported 85.7% with a positive HPV PCR [9].However, in other series HPV was not identified, which may be due to the involvement of as yet unknown subtypes of HPV or the sensitivity of microbiological tests [2]. HPV was not identified in the histological specimens of our patients and we did not request for PCR.

The endoscopic appearance is characteristic but not pathognomonic. It may be confused with glycogenic acanthosis or verrucous carcinoma, a particular type of squamous cell carcinoma. It may present as a sessile or pedunculated polyploid formation, demarcated from adjacent tissue [2].

Most commonly, the size of the papilloma varies between 3 and 12 mm, with an average size of less than 5 mm [10]. Papillomas may be single or multiple, sometimes forming large papillomatoses, and giant forms up to 5 cm have been described [2]. In our series, we also found lesions that were millimetre-sized in the majority of cases, sometimes single or multiple.

Previous studies report variable data regarding the location of oesophageal papilloma. For Bohn et al, the lesions were mainly located in the upper

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oesophagus. However, other studies suggest that the middle and distal oesophagus may also be involved. This could be explained by damage to the mucosa caused by acid reflux [10].

The diagnosis of papilloma is confirmed histologically. It is a benign tumour in which the general architecture of the squamous epithelium is preserved and the basement membrane is intact [2]. In our series, histology was crucial in confirming the diagnosis. Treatment is not yet fully codified, but is essentially endoscopic. Some carry out endoscopic resection using cold forceps [11], a method that has been validated for the majority of our patients, or using hot snare polypectomy or mucosectomy [3,12].

No surveillance has been established. However, some authors suggest that the risk of malignant transformation is very low in case of solitary papilloma, but higher in the case of oesophageal papillomatosis or giant papilloma [2].

Conclusion:

Papilloma of the oesophagus is a rare benign tumour, for which certain factors play a role in its appearance.It has the potential for malignant transformation and the risk increases with the number and size of lesions, hence the importance of identifying them during endoscopy and treating them using the most conservative method possible, often endoscopic resection.

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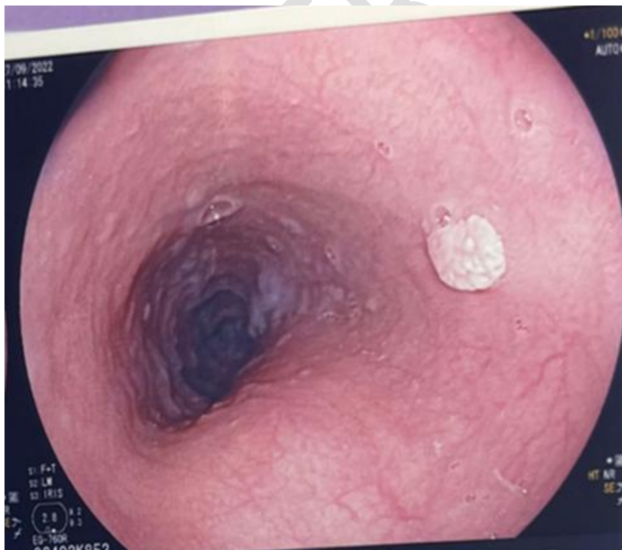

Figure 1: upper endocopy showing an esophageal polyp

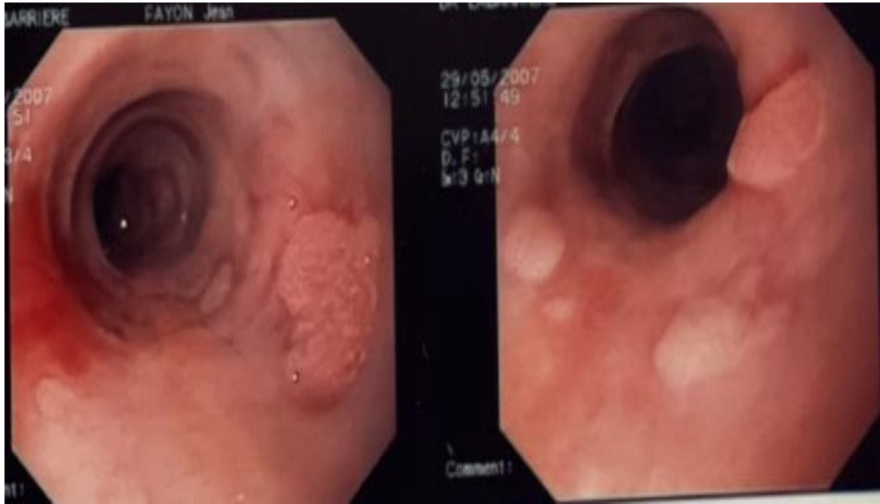

Figure 2 : Several large esophageal lesions

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