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Esophageal squamous papilloma: an esophageal anomaly case report and review

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Introduction:

Esophageal squamous papillomas (ESPs) are rare benign tumors with an estimated prevalence of 0.01-0.43% in the general population.[1] The majority are asymptomatic and an incidental finding on esophagogastroduodenoscopy (EGD), with most being found in the middle to lower third of the esophagus.[1,2,3] A common presenting symptom of ESP is dysphagia refractory to medical treatment. Despite their mostly benign nature, a few cases have been associated with squamous cell carcinoma.[4]

Methods:

A literature search was performed using the PubMed database. Four primary sources were used to examine the characteristics of patients with an ESP. Data from each source was input into Microsoft Excel and used to calculate the mean age, percent of each sex, percent of ESP in each location within the esophagus, and percent of patients that were smokers or alcohol drinkers.

Cases Report and Review Results:

A 42-year-old Caucasian female was referred to gastroenterology for acute on chronic dysphagia for 6 months. The patient had no history of food impactions or reflux disease. Her weight was stable, and laboratory studies were unremarkable. A barium esophagram was completed, notable for a small hiatal hernia and mild reflux in the lower esophagus. Nothing with her clinical history or studies suggested achalasia or an etiology for dysphagia. An upper endoscopy was subsequently scheduled to rule out eosinophilic esophagitis or other intrinsic esophageal pathology to explain her dysphasia. An ESP was found and biopsied.

Table 1 contains the mean age of the patients with ESP in each study. It also contains the total number of each sex found with an ESP. The ages in each study ranged from 43 to 52 years old.

Table 1: Mean Age and Sex of Patients in Each Study

	n	Age	Male	Female
Mosca[1]	9	43.1	4	5
Sablich[2]	35	45	18	17
Talamini[3]	42	45.1	21	21
Jideh[5]	16	52.2	6	10

Italic name = last name of the first author in the respective articles

As shown in table 2 the average overall age was 46 with a essentially equal male to female ratio (48:52 respectively).

Table 2: Mean Age and Percentage of Sex of Patients with ESP

Total Patients	Mean Age	Male (%)	Female (%)
102	46	48	52

Data was combined from references 1,2,3,5 in Table 1.

Table 3 shows the number of ESPs found in each location within the esophagus in 3 of the studies. Overall the middle 1/3 esophagus was the most common location with 45%. However, in Jideh et al study the lower 1/3 esophagus had a higher incidence of ESP. The lower 1/3 was the 2nd most common

location for ESP with 36%. Only 18.9% of ESPs in 3 of the studies were found in the upper third of the esophagus.

Table 3: Location of ESP Within the Esophagus[2,3,5]

	Upper 1/3 esophagus	Middle 1/3 esophagus	Lower 1/3 esophagus
Sablich[2]	7	17	13
Talamini[3]	7	23	12
Jideh[5]	4	3	9
Total	18	43	34
%	18.9	45.3	35.8

Italic name = last name of the first author in the respective articles

Table 4 displays the number of alcohol drinkers in Talamini et al and also the number of smokers in Talamini et al and Jideh et al. 39.7% of the patients in Talamini et al and Jideh et al were smokers.

Table 4: Risk Factors for ESP

	Number of Alcohol Drinkers	Number of Smokers
Talamini[3]	12	16
Jideh[5]	N/A	7
% of patient population	28.6	39.7

Italic name = last name of the first author in the respective articles

Discussion:

On histological examination, ESPs appear as projections of hyperplastic squamous epithelium that cover a fibrovascular core.[1] Neutrophil invasion of the lesion is usually present.[4] Most cases of solitary ESP are asymptomatic and found during an EGD.[1] Biopsy is the gold standard for diagnosis, but the presence of a wart-like solitary lesion raises suspicion for ESP.[4,5] They are found most often in the middle to lower third of the esophagus.[1,5] 81.1% of the patients with an ESP in Sablich et al, Talamini et al, and Jideh et al were found to have one in either the middle or lower third of the esophagus.[2,3,5] The classic patient is middle-aged with GERD. The mean age of the patients in all 4 studies was 46.[1,2,3,5] Being overweight/obese and a smoker are additional risk factors.[5] Presence of hiatal hernia is also a risk factor, but it is thought to primarily be due to the relationship of hiatal hernias to GERD.[5] The etiology of ESP is unclear. The current leading hypothesis is that chronic mucosal irritation from GERD spurs ESP development.[4] Some have proposed that HPV infection could also play a role in the development of ESP, but there is variable prevalence of HPV in these lesions with some studies reporting a prevalence of HPV as low as 0%.[4,5] ESPs that were positive for HPV were found in the middle third to upper third of the esophagus most often rather than the lower third.[4] The ESP's found in the upper 1/3 lacked the neutrophil invasion on histological examination that have been found in the lower third of the esophagus.[4]

Treatment for ESP is usually only used in symptomatic cases because of the low risk of malignancy.[7] Several treatment options include snare polypectomy, cautery, biopsy forceps, radiofrequency ablation, and endoscopic mucosal resection.[5,6,7] Recurrence following removal of the lesion is rare.[2]

A small number of cases have developed into squamous cell carcinoma if untreated.[5] The rare patients with multiple lesions (esophageal squamous papillomatosis) should be treated with esophagectomy because these cases are associated with much higher rates of eventual malignancy.[7] Esophageal squamous papillomatosis lesions are extremely rare with only approximately 12 cases reported in the literature.[6]

Conclusion:

After reviewing the literature of previous cases, we advise ESP to be part of the differential diagnosis in a patient with either GERD or dysphagia that is refractory to medical treatment. Future research still needs to be done on management of esophageal squamous papillomas due to the extremely low prevalence in the population and the uncertain clinicopathological associations.

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