CASE STUDY

# Endoscopic treatment of 135 cases of Plummer-Vinson web: a pilot experience

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The association of postcricoid dysphagia, upper esophageal web(s), and iron deficiency anemia is classically known as Plummer-Vinson syndrome (PVS) or Kelly Paterson syndrome.[1,2] It is a rare entity, particularly in Africa.[1,3,4] However, its recognition is important to identify patients with increased risk of squamous cell carcinoma of the pharynx and esophagus.[1,5-7] Here we report a large series of PVS cases treated endoscopically to evaluate the results of the endoscopic treatment. The epidemiologic, clinical, paraclinical, and evolutive features of these patients are also described.

## METHODS

We conducted a retrospective study over a period of 18 years from September 1993 to December 2011 in our gastroenterology unit. The positive diagnosis of PVS was retained when an upper esophageal web was identified on an upper GI endoscopy. All our patients were treated endoscopically.

Endoscopic treatment was based on dilation performed under oropharyngeal (lidocaine 2% gel) or general (intravenous propofol) anesthesia. Endoscopic dilation was performed using Savary-Gilliard bougies without fluoroscopic guidance. Dilators with a diameter ranging from 9 to 15 mm were selected based on degree of stricture. The most used dilators were 12 to 15 mm. All procedures were carried out by the same operator (Pr A.E.E.). First, a metallic guidewire was inserted under endoscopic control through the web and pushed into the stomach. The endoscope was withdrawn gradually, and the second step was to slip the dilators on the wire with a gradual increase in size without exceeding 3 successive calibers per session. Finally, an endoscopic control was performed to visualize the rupture of the web and to check the integrity of esophageal and gastric mucosa. Esophageal and duodenal biopsy samples were taken in all cases. In the absence of procedural adverse event, patients were allowed a soft food diet and were encouraged to resume a regular diet the following day. Collected data were analyzed statistically using SPSS version 13.0 (SPSS Inc., Chicago, Ill).

*Abbreviations: PVS, Plummer-Vinson syndrome; SCC, squamous cell carcinoma.*

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## RESULTS

We included 135 patients, of which 86.6% were women (sex ratio, 0:15). The mean age was 43 years (range, 14-90).

The mean duration of symptoms before consulting was 5 years and 4 months. Dysphagia was the main symptom observed in 98 patients (5% of cases, n = 133). Grade 0 dysphagia was present in 1.5% (n = 2), grade I in 85% (n = 115), grade II in 7.5% (n = 10), grade III in 4.5% (n = 6), and grade IV in 1.5% (n = 2). Other clinicopathologic features of patients are summarized in Table 1. Of our patients, 83.7% had hypochromic microcytic anemia (n = 113). The mean value of hemoglobin was 8.6 g/dL (±2.1). Iron deficiency anemia was confirmed in 38.5% of patients (n = 52). A decrease in serum ferritin level without anemia was found in 1 patient (.7%). The esophagoscopy showed a cervical esophageal web in all cases. The esophageal web was single in 97.7% of cases (n = 132), and 2 cervical esophageal webs were reported in 2.3% of patients (n = 3). The membrane was circumferential in 93.3% of patients (n = 126) and semilunar in 6.7% (n = 9). Figure 1 shows an example of a circumferential esophageal web. After dilation, esophagoscopy revealed a postcricoid tumor in 2 patients. Associated tumors of the middle and lower thirds of the esophagus were found, respectively, in 2 and 3 patients. Esophageal and/or gastric varices were identified in 7 patients.

All patients underwent an endoscopic treatment. Successful rupture of the web was achieved with Savary-Gilliard

**TABLE 1. Clinical manifestations**

| Symptoms | n | % |
|---|---|---|
| Dysphagia | 133 | 98.5 |
| Tooth loss | 41 | 30 |
| Weight loss | 28 | 20 |
| Goiter | 10 | 7.4 |
| Splenomegaly | 10 | 7.4 |
| Cheilitis | 8 | 6 |
| Glossitis | 8 | 6 |
| Odynophagia | 8 | 6 |
| Edematoascitic syndrome | 5 | 3.7 |
| Chronic diarrhea | 3 | 2.3 |
| Koilonychias | 1 | .7 |
| Brittle nails | 1 | .7 |
| Upper GI bleeding | 1 | .7 |
| Anasarca | 1 | .7 |

dilators in 97% (n = 131) and spontaneously by the endoscope in 3% (n = 4). Figure 2 shows the endoscopic appearance of the web before and after dilation. No immediate adverse event after dilation, such as bleeding or perforation, was registered. Patients with anemia or sideropenia were supplemented by 160 mg of oral ferrous iron daily (n = 114). The mean duration of iron therapy was 6 months; 2.2% of patients (n = 3) required blood transfusion because of the severity of anemia.

During a mean follow-up period of 30.7 months (range, 1-180 months), the outcome was favorable, with a total resolution of dysphagia in 69% of all patients (n = 93); 27.3% of patients (n = 37) had a recurrence of dysphagia and 3.7% had a malignant transformation in the postcricoid region. In the recurrent forms, the relapses occurred in the short-term follow-up period (<6 months) in 5.9% of patients (n = 8), in the medium term (6-12 months) in 5.9% (n = 8), and in the long term (>12 months) in 15.4% (n = 21). One additional dilation session was required in 16.3% of patients, 2 sessions were necessary in 5.2%, and more than 3 additional sessions were indicated in 5.9% (n = 8). Thus, a total of 189 dilation sessions was performed. Figure 3 shows the follow-up of patients having recurrence of dysphagia after endoscopic dilation.

In our series, PVS was complicated and/or associated to many pathologies. Squamous cell carcinoma (SCC) was find concomitant to the diagnosis of PVS in 1.5% of patients (n = 2) and during follow-up in 2.2% (n = 3). The mean interval between the diagnosis of PVS and the diagnosis of SCC was 6 years. Two patients died from cancer progression, and 1 patient was lost to follow-up. Associated cancers of the middle and lower thirds of the esophagus were found, respectively, in 3 and 4 patients (5.2%). The diagnosis of these cancers was concomitant to the diagnosis of PVS in 5 patients and during follow-up in 2 patients. PVS was associated with thyroid disorders in 8.8% of patients (n = 12) and portal hypertension in 5.9% of patients (n = 8). Duodenal biopsy samples revealed typical histologic features of celiac disease in 3.7% of patients (n = 5). Figure 4 summarizes all pathologies associated with PVS in our series.

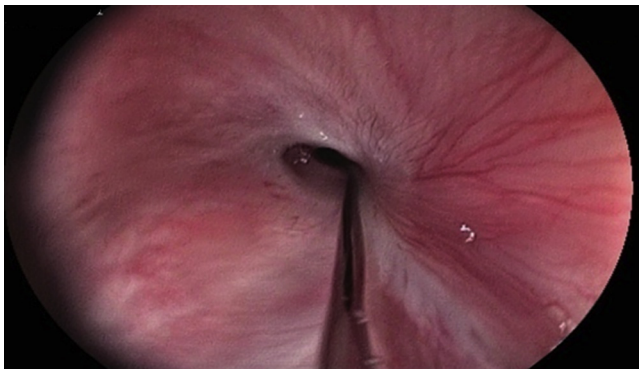

**Figure 1.** Circumferential upper esophageal web.

## DISCUSSION

Until the 1960s, PVS was frequently described in the Caucasian population of the northern countries of Europe, especially in Scandinavia.[1,3] Currently, it is a very rare entity in the world, especially in Africa,[1,8] and exact data about its incidence and prevalence are lacking.[1] Most publications are case reports and short series of patients recruited over several years and from various countries.[1-4,7-11] To our knowledge, our series can be considered among the largest series of PVS published in the medical literature.

The pathogenesis of this syndrome is unknown, but iron deficiency appears to play a major role. Other etiologic factors, including malnutrition, genetic predisposition, or even autoimmune processes, have been proposed.[1]

In comparison with other studies, the mean age of our patients (43 years) was slightly lower than the average of 47 years described in all case reports published in the English medical literature between 1999 and 2005.[1] Other series show comparable results in African populations (Ben Gamra et al,[3] 43 years; Dia et al,[4] 37 years) and Asian patients (Okamura et al,[10] 45.5 years). Analysis of these data confirm that this syndrome is predominantly prevalent in the middle-aged adult and that PVS affects mainly women, which account for 86.6% of the patients in our study.