

# Plummer-Vinson Syndrome in Senegal: A Report of 219 Cases at the Aristide Le Dantec Hospital in Dakar

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## Abstract

**Introduction:** Plummer-Vinson syndrome (PVS), also known as Kelly-Pater-son syndrome, is a rare condition classically defined by the combination of dysphagia, iron-deficiency anemia, and a mucosal web of the cervical esophagus. The objective of our study was to describe the epidemiological, clinical, paraclinical, therapeutic, and outcome characteristics of PVS in our setting, and to identify factors associated with web resolution, recurrence, and the presence of gastrointestinal cancer. **Patients and Methods:** We conducted a retrospective, descriptive, and analytical study from 1 January 2005 to 31 December 2022 at the digestive endoscopy center of the Hepato-Gastroenterology Department of Aristide Le Dantec Hospital in Dakar. We included all patients in whom esophagogastroduodenoscopy revealed a mucosal web of the cervical esophagus compatible with PVS. **Results:** The prevalence of SPV was 1.04%. The mean age was 36 years [13 - 96 years], and the sex ratio was 0.10. Dysphagia was present in all patients. Hypochromic microcytic anemia was found in 54% of patients. On endoscopy, all patients had a stenosing membranous ring in the cervical esophagus. At the time of diagnosis, 8 patients had esophageal cancer and two had gastric cancer. Endoscopic dilation was performed in 89% of patients. At 9 months, 16 patients had persistent dysphagia, 28 had recurrence of the ring, and 7 had a refractory stricture; one case of high-grade dysplasia was observed after 10 years of follow-up. In univariate analysis, a high location of dysphagia and a normal serum iron level were associated with the disappearance of the ring, whereas anemia was associated

with recurrence. Male sex, age > 35 years, low socioeconomic status, smoking, and both high and low dysphagia were associated with the presence of gastrointestinal cancer. **Conclusion:** In our setting, SPV primarily affects young women. Despite the effectiveness of endoscopic dilation, the occurrence of recurrences and neoplastic lesions justifies prolonged follow-up.

### Keywords

Plummer-Vinson Syndrome, Dysphagia, Iron-Deficiency Anemia, Esophageal Web, Endoscopic Dilation, Esophageal Cancer

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## 1. Introduction

Plummer-Vinson syndrome (PVS), also known as Kelly-Paterson syndrome or sideropenic dysphagia, is classically defined by the association of upper dysphagia, iron-deficiency anemia, and an esophageal web of the cervical esophagus [1]. Formerly more common in Western countries, it has become exceptionally rare there, but continues to be observed in developing countries, particularly in Africa [2]-[6]. Its pathophysiology remains incompletely understood, although iron deficiency appears to play a central role [1]. Clinical manifestations are dominated by dysphagia and symptoms related to iron deficiency. Treatment is based on iron supplementation and endoscopic dilation. PVS is also associated with an increased risk of cancer of the hypopharynx and the upper gastrointestinal tract, particularly the esophagus [7]. The objective of our study was to describe the epidemiological, clinical, paraclinical, therapeutic, and outcome characteristics of PVS in our setting, and to identify factors associated with web resolution, recurrence, and the presence of gastrointestinal cancer.

## 2. Patients and Methods

We conducted a retrospective, descriptive, and analytical study over an 18-year period, from January 1, 2005 to December 31, 2022. The study was carried out at the digestive endoscopy center of Aristide Le Dantec Hospital.

The study population comprised all patients who underwent upper gastrointestinal endoscopy during the study period. We included all patients presenting with dysphagia associated with a cervical esophageal web identified on upper gastrointestinal endoscopy and compatible with Plummer-Vinson syndrome. Patients with unusable medical records were excluded.

Data were collected from medical records and endoscopy reports using a standardized data collection form. The variables examined included sociodemographic characteristics, medical history, and clinical, laboratory, endoscopic, therapeutic, and outcome data.

Dysphagia was classified according to its location as high, low, or high and low. Socioeconomic status was assessed using occupation, living conditions, and socioeconomic information available in the medical records, and classified as low,

middle, or high.

Biopsies were performed for all lesions suspicious for malignancy, and all cancers included in the study were histologically confirmed.

Recurrence was defined as the inability to maintain a satisfactory luminal diameter for 4 weeks after achieving a diameter of 14 mm or as the endoscopic re-appearance of the ring after its initial disappearance.

Refractory stricture was defined as the inability to achieve an esophageal diameter  $\geq 14$  mm after five dilation sessions performed at 4-week intervals.

Data were entered using SphinxPlus<sup>2</sup> software (version 5) and then analyzed with SPSS Statistics (version 25). Qualitative variables were expressed as counts and percentages. Quantitative variables were reported as mean  $\pm$  standard deviation or as median, depending on their distribution. Comparisons were performed using the chi-square test or Fisher's exact test, as appropriate. The threshold for statistical significance was set at  $p < 0.05$ .

### 3. Ethical Considerations

Our study was retrospective and relied exclusively on the use of pre-existing medical records. It did not expose patients to any additional risk and did not involve the performance of additional examinations or the prospective collection of new data. The information collected was processed in strict accordance with medical confidentiality and privacy requirements.

### 4. Results

During the study period, 21,850 upper gastrointestinal endoscopies were performed. Among 228 patients initially identified as having Plummer-Vinson syndrome, 9 patients were excluded due to unusable medical records. The analysis therefore included 219 patients, corresponding to an in-hospital prevalence of 1.04%.

The mean age was 36 years [13 - 96 years], with a predominance of the 20 - 30 age group (33.3%). There were 199 women (90.9%), corresponding to a sex ratio of 0.10. Most patients were from the Dakar region (63.5%). Socioeconomic status was assessed as intermediate in 14% of patients.

A history of bleeding was identified in 53 patients (24.2%), most often chronic (23.7%) and of gynecological origin (22.8%). Comorbidities were rare, mainly hypertension (1.8%) and type 2 diabetes (1.4%). Active smoking was noted in 2 patients (1.0%). Oral herbal medicine use was reported in 10 patients (4.6%), and regular clay consumption in 22 patients (10.0%).

SPV was diagnosed in all patients during the evaluation of dysphagia. The mean time to diagnosis was 25 months [1 month - 30 years]. Dysphagia was present in all cases (100.0%). It was proximal in 95.4% of cases, distal in 1.0%, and both proximal and distal in 3.7%. It was most often persistent (81.3%) and predominantly affected solids (73.5%). The main associated signs were dizziness (25.1%) and headache (15.1%). The most frequent general signs were mucosal pallor (47.5%) and asthenia (47.0%). General condition was preserved in most patients (WHO 0:

76.3%). Stigmata of iron deficiency was mainly angular cheilitis (63.0%) and koilonychia (10.5%).

The patients' sociodemographic and clinical characteristics are presented in **Table 1**.

**Table 1.** Socio-demographic and clinical characteristics of patients.

	Sample size (n)	Percentage (%)
<b>Sex</b>		
Women	199	90.9
Men	20	9.1
<b>Age: Mean [range] (years)</b>		
	36 years [13 - 96 years]	
<b>History of bleeding</b>		
Polymenorrhea	27	12.3
Metrorrhagia	21	9.6
Fibroid	2	0.9
Rectal bleeding	2	0.9
Hematemesis	1	0.5
<b>Functional signs</b>		
Dysphagia	219	100.0
Dizziness	55	25.1
Headaches	33	15.1
Pyrosis	2	0.9
Epigastric pain	2	0.9
Hypersialorrhea	1	0.5
<b>General signs</b>		
Pallor of the mucous membranes	104	47.5
Asthenia	103	47.0
Weight loss	52	23.7
Anorexia	16	7.0
Malnutrition	1	0.5
<b>Physical signs</b>		
Angular cheilitis	138	63.0
Koilonychia	23	10.5
Fine, dry hair	8	3.7
Leukonychia	6	2.7
Glossitis	4	1.8
Alopecia	2	0.9
Brittle nails	1	0.5

A complete blood count was performed in all patients. The mean hemoglobin level was 10.58 g/dL [4 - 14 g/dL], and microcytic hypochromic anemia was observed in 118 patients (54.0%). Iron studies were available for 142 patients (64.8%). The mean serum ferritin level was 39.15 ng/mL [0.3 - 766.7 ng/mL], with hypoferritinemia in 70/142 patients (49.3%). The mean serum iron level was 42.82 µg/dL [0.05 - 167.32 µg/dL], with hyposideremia in 103/142 patients (72.5%). Isolated iron deficiency was noted in 2/142 patients (1.4%).

On oesophagogastroduodenoscopy, all patients had a stenosing membranous ring of the cervical oesophagus, circumferential in 210 cases (95.9%). Associated lesions were observed in 158 patients (72.1%), predominantly gastritis (16.9%), cardiac incompetence (16.4%), and a Schatzki ring (14.2%). Neoplastic lesions were also identified in 10 patients (4.6%), involving the oesophagus in 8 cases and the stomach in 2 cases. The lesions observed on oesophagogastroduodenoscopy are summarized in **Table 2**.

**Table 2.** Lesions observed on esophagogastroduodenal endoscopy.

Endoscopic lesions	Effective	Percentage (%)
<b>Plummer-Vinson ring</b>	219	100.0
<b>Associated lesions</b>	158	72.1
Gastritis	37	16.9
Cardiac incontinence	36	16.4
Schatzki ring	31	14.2
Esophagitis	26	11.9
Hiatal hernia	10	4.6
Esophageal tumor	8	3.7
Polyp	7	3.2
Gastric ulcer	3	1.4
Gastric tumor	2	0.9

An esophagogastroduodenal transit study was performed in 189 patients (86.3%) and demonstrated an annular stenosis in 185/189 patients (97.9%), most frequently located in the cervical esophagus (98.9%).

Histopathological results were available for 34 patients. They revealed esophageal squamous cell carcinoma in 5 cases, adenocarcinoma of the lower third of the esophagus in 3 cases, and gastric adenocarcinoma in 2 cases. In addition, 21 patients had chronic atrophic gastritis, associated with *Helicobacter pylori* in 12 cases.

Oral iron supplementation, at a dose of 160 mg/day for 2 to 6 months, had been prescribed in 125 patients (57.1%).

At the initial endoscopy, ring collapse was observed in 9 patients (4.1%). Endoscopic dilation using Savary-Gilliard bougies, under diazepam sedation, was performed in 195 patients (89.0%). The mean number of sessions was 2 [1 - 19 ses-

sions], with a mean of 3 bougies per session [2 - 6 bougies] and a mean diameter of 13 mm [7 - 15 mm].

At 9 months of follow-up, 27 patients were lost to follow-up, including 15 from the time of diagnosis. Among the patients who were reassessed, 16 had persistent dysphagia, 3 had anemia, 4 had low serum iron, and 1 had low ferritin. Ring recurrence was observed in 28 cases, and refractory stenosis in 7 cases.

Univariate analysis showed that a high location of dysphagia ( $p = 0.045$ ) and a normal serum iron level ( $p < 0.001$ ) were significantly associated with resolution of the esophageal ring after endoscopic treatment. The presence of anemia appeared to be a significant risk factor for ring recurrence (OR = 6.19; 95% CI: 2.904 - 24.022;  $p = 0.043$ ). Factors associated with the presence of a digestive cancer were male sex, age > 35 years, low socioeconomic status, smoking, and both high and low dysphagia (Table 3).

**Table 3.** Factors associated with the presence of digestive cancer.

Variable	Reference category	OR	95% CI	p-value
Male sex	Female sex	8.042	[2.056 - 31.45[	0.0005
Age >35 years	≤35 years	4.36	[1.904 - 21.022[	0.047
Low socioeconomic status	Middle/high socioeconomic status	9.66	[1.75 - 125]	0.046
Smoking	No-smokers	23.11	[1.336 - 399.94[	0.002
Upper and lower dysphagia	High dysphagia	-	-	0.000

## 5. Discussion

In the first half of the 20<sup>th</sup> century, SPV appeared to be common in Western countries, particularly among young women. With improvements in nutritional status and the development of maternal health programs, it has become rare there, with only a few published observations [1]. However, it is still observed in Africa, where several authors have reported case series [2]-[6]. In our study, 219 patients had SPV, corresponding to a prevalence of 1.04%.

It mainly affected young patients, with a mean age of 36 years (range, 13 - 96 years) and a predominance in the 20 - 30-year age group, consistent with series previously reported in Senegal [3] [5]. However, our series confirms that SPV can be observed at any age, including in children, adolescents, and older adults, as already described in the literature [8] [9]. The marked female predominance observed is consistent with the classical data reported in the literature [1] [2] [4] [5]. This is usually attributed to the central role of iron deficiency, promoted by menstruation, repeated pregnancies, and breastfeeding [1].

Clinically, dysphagia was the consistent presenting symptom, observed in all patients, as in most published series [4] [10] [11]. Signs of iron deficiency were frequent, predominated in our study by angular cheilitis, mucosal pallor, asthenia, and, more rarely, koilonychia.

From a biological standpoint, hypochromic microcytic anemia was found in 54.0% of patients, and iron deficiency in 72.5%. The frequency of anemia in our study is close to that reported by Hefaiiedh [8] (53%) but lower than those observed by Chtourou [11], Ben Gamra [10], and Dia [4], who reported 100%, 89.4%, and 79%, respectively. The observed differences may be explained by heterogeneity in the study populations, as well as by the fact that some of our patients were already receiving iron therapy at the time of diagnosis. Moreover, the absence of overt anemia or frank iron deficiency in some patients suggests that iron deficiency, although central to SPV, is not sufficient on its own to explain the pathophysiology. Other mechanisms, notably nutritional, genetic, or autoimmune, may also be involved [1]. In our series, the possible causes of iron deficiency were dominated by chronic gynecological bleeding (22.8%), far ahead of gastrointestinal causes. Geophagia was reported in 10.0% of cases, and one patient had pernicious anemia, which strengthens the hypothesis of a multifactorial iron deficiency.

The mean diagnostic delay was 25 months. Although this interval remains long, it appears shorter than that reported in some earlier series, notably that of Dia [4], in which it reached 8 years. Several factors may account for these diagnostic delays: limited access to specialist care, the slowly progressive course of symptoms, trivialization of initial dysphagia, and socioeconomic constraints. The relative shortening of the diagnostic delay observed in our series may reflect greater practitioner awareness of the disease as well as broader access to upper gastrointestinal endoscopy.

Upper gastrointestinal endoscopy has confirmed its central role in diagnosis by demonstrating a cervical membranous ring in all patients, as reported in the series by Bakari [2], Dia [4], and Hefaiiedh [8]. This ring was stenotic in all cases and most often circumferential. Endoscopy also made it possible to identify numerous associated lesions, including gastritis, cardiac incompetence, Schatzki rings, and esophagitis. It also revealed digestive tumors in 10 patients, highlighting the value of this examination not only for establishing the diagnosis but also for screening for potentially serious associated lesions.

Oesophagogastroduodenal transit, performed in 86.3% of patients, also identified an annular cervical stenosis in most cases. Although less specific than endoscopy in our practice, it remains descriptively useful for the morphological assessment of stenosis when it cannot be traversed.

Management relied primarily on endoscopic dilation using Savary-Gilliard bougies, performed in 89.0% of cases, combined with iron supplementation in 57.1% of patients. Spontaneous disruption of the ring by the endoscope was observed in 4.1% of cases. These results are consistent with those in the literature [4] [5], confirming the central role of endoscopic dilation combined with correction of iron deficiency in the management of SPV.

In our study, 28 ring recurrences were observed during follow-up. Recurrence is also reported in the literature, with a frequency that varies across series [2] [3] [8]. Anemia was associated with recurrence, whereas a normal serum iron level

was associated with resolution of the ring after treatment. These findings suggest that more severe iron deficiency may be linked to a less favorable course and underscore the importance of adequate correction of iron deficiency, control of the etiological factor, and prolonged follow-up after dilation.

The initial endoscopy identified 10 digestive cancers, including 8 esophageal cancers and 2 gastric cancers, representing 4.6% of cases at the time of diagnosis. Associations between PVS and neoplastic lesions have also been reported in the literature, notably by Dia [3], Kadiri [5], and Patil [12]. Over an 8-year period, Bassène [13] reported 7 cases of Plummer-Vinson rings associated with esophageal cancer. In addition, one case of high-grade dysplasia was observed during follow-up. Collectively, these findings reinforce the preneoplastic nature of PVS and justify careful endoscopic surveillance. Although there is no consensus regarding its modalities, some authors propose annual surveillance [12] [14]. In our study, male sex, age > 35 years, low socioeconomic status, high and low dysphagia, and smoking were associated with the presence of digestive cancer at diagnosis. However, these results should be interpreted with caution because of the limited number of cancer cases and the rarity of some exposures. Further studies including larger populations are needed to confirm these associations.

The observation of an association with Biermer's disease, although rare, deserves emphasis. It is consistent with other cases reported in the literature [15] [16] and suggests that, beyond iron deficiency alone, autoimmune mechanisms may also contribute to the pathophysiology of SPV.

## 6. Study Limitations

This study has some limitations. Its retrospective and single-center design may have introduced selection bias and missing data, particularly regarding iron studies. In addition, some patients were lost to follow-up, limiting long-term outcome assessment. Finally, the statistical analyses were based solely on univariate analyses, which does not allow exclusion of potential confounding factors.

## 7. Conclusion

Plummer-Vinson syndrome remains a rare condition in our setting, occurring predominantly in young women and most often presenting with dysphagia. Upper gastrointestinal endoscopy plays a central role in diagnosis, assessment of associated lesions, and treatment. Despite the overall effectiveness of endoscopic dilation combined with correction of iron deficiency, the occurrence of recurrences and neoplastic lesions warrants early management and long-term surveillance.

## Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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