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FREQUENCY OF PLUMER-VERSION SYNDROME IN PATIENTS PRESENTING WITH DYSPHAGIA IN ENT DEPARTMENT NISHTAR HOSPITAL

Dr Sabin Sajida[1], Dr Deepak Rai[2*], Dr Afshan Qayum[3], Dr Mahreen Tahir[4], Dr Umera Iftikhar[5], Dr Sanaullah Bhatti[6]

[1]Senior Registrar, Bakhtawar Amin Teaching Trust Hospital, Pakistan
Email: sabeensajida@yahoo.com
[2*]Senior Registrar, Dow University of Health Science (Civil Hospital) Karachi, Pakistan
Email: dr_deepakrai@hotmail.com
[3]Senior Registrar, Sheikh Zayed Medical College, Rahim Yar Khan, Pakistan
Email: Fishqayum22@gmail.com
[4]Senior Registrar, Bakhtawar Amin Teaching Trust Hospital, Email: drmahreen8@gmail .com
[5]Senior Registrar ENT, Mohi Ud Din Teaching Hospital Mirpur Azad Jamu Kashmir
Email: Dr.umeraiftikhar@gmail.com
[6]Assistant Professor, Bakhtawar Amin Teaching Trust Hospital, Pakistan
Email: sanaullahbhatti187@gmail.com

***Corresponding author:** Dr Deepak Rai
*Email: dr_deepakrai@hotmail.com

ABSTRACT

Introduction: The Plummer-Vinson syndrome (PVS) is an uncommon disorder characterized by iron deficiency anaemia, oesophagal webs and dysphagia, frequently found in Pakistan and other developing countries where nutritional deficiency may be the cause.

Objective: The frequency of Plummer-Vinson syndrome was to be measured in patients who presented with dysphagia to the ENT Department, Nishtar Hospital, Multan, Pakistan.

Materials and Method: The study was descriptive and cross-sectional, conducted from September 2018 to March 2019 in Nishtar Hospital, Multan. Patients with malignancy, neurogenic causes or foreign body-related dysphagia were excluded. The study was performed on 97 patients aged 15 to 45 years. Data collected were ENT history, physical examination, iron studies and barium swallow X-ray and were analyzed with SPSS 20.

Results: Of 97 patients, 10 (10.31%) were diagnosed with PVS. Mean age was 31.85 ± 7.93 years; 61.86% were male. No significant association was found with age, gender, or symptom duration ($p>0.05$).

Conclusion: PVS is a notable cause of dysphagia in Pakistan, requiring early diagnosis to reduce morbidity and cancer risk.

Keywords: Plummer-Vinson syndrome, dysphagia, iron deficiency anemia, esophageal webs.

INTRODUCTION

Plummer-Vinson syndrome (PVS) or Paterson-Brown-Kelly syndrome is a rare clinical condition characterized by a triad of iron deficiency anaemia, oesophagal webs and dysphagia. This represents a syndrome that almost exclusively afflicts middle-aged women and has generated interest owing to its association with dysphagia and possible relationship with upper gastrointestinal malignancies. The condition is named after the United States physicians Henry Stanley Plummer and Porter Paisley Vinson, but in the United Kingdom, after Paterson and Brown Kelly, indicating a transcontinental popularity of the condition (1). The syndrome is characterized by difficulty in swallowing solids, which may progress to difficulties with liquids and symptoms of iron deficiency such as fatigue, pallor and koilonychia (spoon-shaped nails) (2). Despite a dramatic decrease in its prevalence in developed nations as a consequence of improved nutrition and iron supplementation, PVS remains a major problem in developing areas like parts of South Asia and Africa, where iron deficiency is common (3).

The pathogenesis of PVS is not understood, and many theories have been generated (iron deficiency, genetic predisposition or autoimmune factors). Iron deficiency appears to be a central etiological factor that causes mucosal atrophy and the production of oesophagal webs, which physically obstruct the oesophagus and cause dysphagia (4). Myasthenic changes in swallowing muscles may be secondary to the depletion of iron-dependent enzymes (5). While the exact mechanism is unclear, iron deficiency does not always correlate with oesophagal webs, and webs can arise in non–anaemic patients (6). Some cases of PVS have been linked with autoimmune conditions such as celiac disease, rheumatoid arthritis and thyroiditis (7). For instance, there have been studies documenting the coexistence of PVS and celiac disease, thought to have developed secondary to chronic malabsorption and iron deficiency (8).

PVS presents clinically with dysphagia, often intermittent and to solids only felt in the throat, secondary to the proximal location of oesophagal webs (9). Such iron deficiency is accompanied by additional manifestations such as angular cheilitis, glossitis, pallor and, more rarely, splenomegaly or dementia (10). PVS is rare in the pediatric population, has been documented and also needs to be considered across all age groups (11). Laboratory tests such as serum ferritin, haemoglobin and total iron binding capacity reveals iron deficiency anaemia, while oesophagal webs are discovered by barium swallow X-ray or upper gastrointestinal endoscopy (12). Esophagram barium is highly sensitive, revealing webs as thin projections off the anterior oesophagal wall, often best seen on lateral views.

In addition, the implications of PVS are beyond its symptoms, given its association with a greater risk of squamous cell carcinoma of the hypopharynx and oesophagus, with frequencies reported between 4 – 16% in older studies (13). The importance of early diagnosis and long-term monitoring of these affected patients is reflected in this malignancy risk. Iron supplementation for anaemia may be adequate to resolve dysphagia in cases with mild webs, while more obstructing webs usually require mechanical dilation, usually by endoscopy. Advanced interventions such as balloon dilation or laser therapy may also be used to break webs, although with high success rates (14). Despite better nutrition, iron fortification in the diet and increased access to healthcare, PVS prevalence has declined in developed countries, but its persistence in places such as Pakistan requires targeted studies to determine its burden (3).

Measurement of the frequency of Plummer-Vinson syndrome is the objective of this study among patients presenting with dysphagia at the ENT Department of Nishtar Hospital Multan Pakistan. Since data on the occurrence of PVS in developing countries remains scarce, this research intends to offer some insights into the prevalence of PVS in a designated clinical setting as a way of understanding its epidemiology and providing pieces of useful information necessary for ideal diagnostic and management approaches to the condition (4). This study fills a critical gap in regional data by focusing on a cohort of patients with dysphagia and may guide clinical practice as well as show the need for PVS screening in similar populations. These findings may also help shape public health approaches to reduce iron deficiency and its sequelae in resource-poor settings.

Objective: The aim of this study was to determine the frequency of Plummer-Vinson syndrome in patients with dysphagia who presented to the ENT Department of Nishtar Hospital, Multan, Pakistan, from September 2018 to March 2019.

MATERIALS AND METHODS

Design: cross-sectional study.

Study setting: The study was carried out at the Department of Otorhinolaryngology, Nishtar Hospital, Multan, Pakistan, a tertiary care facility with a specialized ENT unit.

Duration: The study spanned six months, from September 14, 2018, to March 13, 2019.

Inclusion Criteria: Inclusion criteria were patients aged 15 to 45 years old, both males and females, who came to the ENT outpatient department with dysphagia. Clinical history of dysphagia was confirmed by indirect laryngoscopy, which revealed pooling of saliva.

Exclusion Criteria: Patients with dysphagia due to oropharyngeal and hypopharyngeal malignancies, neurogenic dysphagias (predominantly affecting liquids) or radiologically identified foreign bodies on soft tissue neck lateral view and causing acute onset of dysphagia were excluded.

Methods

Particular proforma was used for study recording after obtaining approval from the ethical committee at Nishtar Hospital, Multan. The patients meeting the inclusion criteria were selected from the Patients Department of otorhinolaryngology. However, informed consent was obtained after explaining all of the study's risks and benefits. Detailed ENT history followed by general physical and ENT examination was taken. Serum iron, serum ferritin, total iron-binding capacity and barium swallow X-ray to detect oesophagal webs were investigated as routine. SPSS version 20 was used to analyze data. Numerical variables were described by mean, and standard deviation, and categorical variables were described by frequencies and percentages (gender, PVS diagnosis). All analyses were stratified for the effect modifiers age, gender, and symptom duration, and the impact on outcomes was tested with the chi-square test ($p<0.05$ statistically significant).

RESULTS

The study included 97 patients presenting with dysphagia at the ENT Department of Nishtar Hospital, Multan, from September 14, 2018, to March 13, 2019. The age range of participants was 15 to 45 years, with a mean age of 31.85 ± 7.93 years. The majority, 55 patients (56.70%), were aged between 31 and 45 years, while 42 (43.30%) were between 15 and 30 years. This distribution highlights a higher prevalence of dysphagia among older patients within the studied age group, consistent with patterns observed in Plummer-Vinson syndrome (PVS) literature (1,7).

Table I: Age Distribution of Patients (n=97)

Age Group (Years)	Frequency	Percentage (%)
15–30	42	43.30
31–45	55	56.70
Mean ± SD = 31.85 ± 7.93 years		

Of the 97 patients, 60 (61.86%) were male, and 37 (38.14%) were female, resulting in a male-to-female ratio of 1.6:1. This gender distribution suggests a slight male predominance, which contrasts with some studies reporting higher female prevalence in PVS, particularly in Western populations (10,11). Dysphagia symptoms in most cases present rather chronically with a mean duration of 3.46 ± 1.34 months, consistent with the progressive nature of PVS-related dysphagia (8, 12).

Table II: Gender Distribution of Patients (n=97)

Gender	Frequency	Percentage (%)
Male	60	61.86
Female	37	38.14

The frequency of Plummer-Vinson syndrome in patients with dysphagia was 10 (10.31%). The prevalence here is higher than some reported figures, like in a study in Peshawar where 1 out of 20 dysphagia patients had PVS (10). The diagnosis was made by clinical assessment, iron studies (in particular serum iron, ferritin and total iron binding capacity) and a barium swallow X-ray for oesophagal webs. The importance of PVS as a major cause of dysphagia in this population is underscored by these findings and a developing country setting where iron deficiency is prevalent (3,6).

Table III: Duration of Symptoms (n=97)

Duration (Months)	Frequency	Percentage (%)
1–3	52	53.61
>3–6	45	46.39
Mean ± SD = 3.46 ± 1.34 months		

Analyses stratified by age groups ($p = 0.07$), gender ($p = 0.12$), or symptom duration ($p = 0.09$) using the chi-square test did not reveal any significant association between PVS frequency and these features. These results highlight the need to screen for PVS in all dysphagia patients independent of demographic variables to identify PVS in a timely manner.

DISCUSSION

This study of 97 patients conducted at the ENT department, Nishtar Hospital Multan Pakistan, showed the incidence of Plummer Vinson Syndrome 10.31 % (10 out of 97 patients) in dysphagia. The prevalence of PVS in both groups is greater than that reported by a study from Peshawar, which showed a 5% PVS rate among dysphagia patients (10). This observed difference may be caused by regional differences in nutritious status, especially in iron deficiency, which is a factor in the aetiology of PVS (1). The age range of the study (15 to 45 years, mean 31.85 ± 7.93 years) is typical for PVS, even though the classical description of the syndrome is in middle-aged women, mainly in the fifth decade (7). In this study, the majority of patients were 31–45 years old with young presentation (56.70%), maybe due to a high burden of iron deficiency in developing countries like Pakistan (3). Historical data show a predominance of the female gender (90% in older Scandinavian studies), while the current data show a male predominance (ratio 1.6:1), suggesting generalized differences in gender distribution in modern context or regional differences (11).

The pathophysiology of PVS is speculative, with iron deficiency as the most widely held etiological factor. Mucosal atrophy and the development of oesophagal webs, which physically obstruct the oesophagus, resulting in dysphagia, can result from iron deficiency (4). There is evidence that iron supplementation can resolve dysphagia in some patients, particularly in those with less obstructive webs (5). However, PVS is not present in many iron-deficient populations, e.g. in eastern and central Africa, casting doubt on its sole occurrence in iron deficiencies (6). The diagnostic protocols for the current study included serum iron, ferritin, total iron binding capacity and barium swallow X-ray, which independently confirmed iron deficiency anaemia and oesophagal webs, which were in agreement with classical diagnostic protocols (12). Barium esophagogram, the most sensitive test for oesophagal webs, was the test crucial in demonstrating the characteristic thin, crescent-shaped projections in the proximal oesophagus (13). The finding of a 10.31% prevalence of PVS indicates the need for routine screening for PVS in dysphagia patients, particularly in populations in which nutritional deficiencies are prevalent.

There are also autoimmune factors implicated in PVS, including associations with celiac disease, rheumatoid arthritis, and thyroiditis (7). PVS as a presenting feature of celiac disease was reported in a case report from Romania, and malabsorption-related iron deficiency is thought to play a role in the development of the syndrome (4). A similarly discordant case series in Tunisia reported autoimmune conditions in some PVS patients and reinforced the potential for autoimmune (8). While the study did not test specifically for antibodies against autoimmune markers, the high prevalence of PVS is in a region where nutritional challenges (and thus iron deficiency) are known, so autoimmunity is not likely to be the driving force. A second proposed factor is genetic predisposition, where several familial cases have been reported but were not investigated in the current study (9). PVS was rare in pediatric populations since no patients under 15 years were included, similar to a case report of a rare pediatric presentation (2).

Beyond dysphagia, PVS has clinical significance because of the association with the increased risk of hypopharyngeal and oesophagal squamous cell carcinoma, which has a historical incidence of malignancy risk of 4–16% (14). However, the risk associated with this condition is that it requires long-term monitoring, with regular endoscopic evaluations in particular within the first few years after diagnosis (1). The current study's findings support the need for heightened vigilance in dysphagia patients in order to help identify PVS early for timely intervention and cancer surveillance. This study did not address any of the treatment strategies observed in the literature (iron supplementation and endoscopic dilation), which are key in the management of PVS. A study of PVS demonstrated that endoscopic dilation has an efficacy rate of 94% for complete response to dysphagia (14). Advanced techniques such as balloon dilation or laser therapy are successful in cases in which webs are highly obstructive (5).

The high prevalence in this study compared to Western populations, in which PVS is a rare problem due to improved nutrition and iron fortification, emphasizes the continuing problem of iron deficiency in Pakistan (10). Consistent with these findings is a case report of a Pakistani woman with PVS, and the syndrome is relevant in South Asian contexts. Chronic presentation with a mean symptom duration of 3.46 ± 1.34 months is consistent with reports that patients frequently have prolonged tolerance of progressive dysphagia until they seek medical attention (12). The delay may make morbidity such as malnutrition and weight loss worse for those with more severe dysphagia (9). A lack of significant associations of PVS frequency with gender, age and symptom duration ($p>0.05$) are consistent with these results and further strengthens the argument that PVS should not be excluded and should be considered in all dysphagia cases, as currently recommended by comprehensive diagnostic workup.

This study holds practical implications for clinical practice in Pakistan, where healthcare resources are so limited. Routine screening for iron deficiency and oesophagal webs in dysphagia patients would improve diagnostic rates and the occurrence of complications, including aspiration pneumonia or malignancy (11). PVS is found to be related to thyroid–cardiac diseases, such as heart failure, complicating management, necessitating a multidisciplinary approach (13). Customized treatment plans are required because of pharmacotherapy problems among HIV patients with PVS. The main goal of this study was to demonstrate that early detection and control or treatment of PVS can prevent its morbidity as well as possible malignant transformation, particularly in areas where the prevalence of iron deficiency is high.

CONCLUSION

The study was carried out on patients in the ENT department, Nishtar Hospital Multan, Pakistan, from September 2018 to March 2019 that established Plummer Vinson Syndrome (PVS) with the frequency of 10.31% in patients' population among 97 dysphagic patients admitted to ENT Nishtar Hospital. This would indicate a higher occurrence of PVS than some regional reports and indicates PVS likely represents a major cause of dysphagia, a high prevalence area for iron deficiency. However, routine screening for PVS by iron studies and barium swallow X-ray should be essential in a country like Pakistan, facing nutritional deficiency and with definite potential for such problems. PVS carries an increased risk of hypopharyngeal and oesophagal cancers, and it is important to have an early diagnosis and to follow up with regular endoscopic surveillance. Clinicians should prioritise early

evaluation in dysphagia patients to enable early iron supplementation and early endoscopic interventions, which may improve outcomes and reduce morbidity. These (PVS) have been mitigated to some extent by enhanced awareness and diagnostic protocols in such populations.

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