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Case Report

Dentistry Section

Management of Plummer-vinson Syndrome: A Report of Two Cases

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ABSTRACT

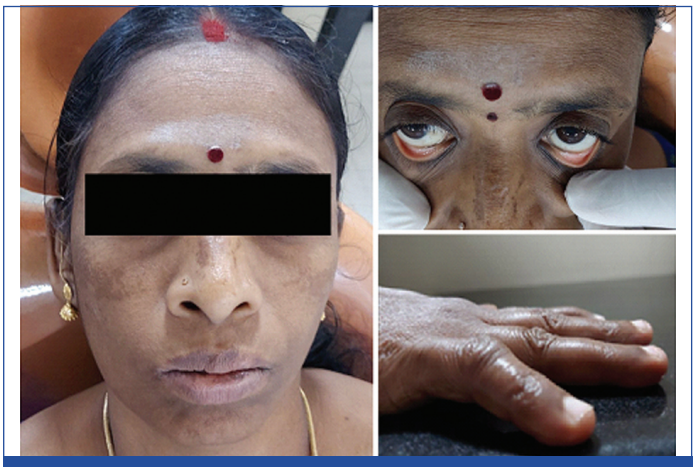
Plummer-Vinson Syndrome (PVS), also referred to as Paterson-Kelly syndrome, is a complication of Iron Deficiency Anaemia (IDA) that is associated with the formation of oesophageal webs and dysphagia. The oesophageal webs are thin, membranous structures that partially obstruct the upper oesophagus, hindering the passage of food. PVS occurs more frequently in middle-aged women and is linked to an increased risk of squamous cell carcinoma of the pharynx and oesophagus. Due to the high risk of malignant transformation, PVS has been grouped under the category of Oral Potentially Malignant Disorders (OPMD). Identification of the disease, early intervention, and long-term follow-up are essential to prevent complications associated with PVS. A thorough history and clinical examination to evaluate possible etiologic causes, haematological evaluation of the blood count and erythrocyte morphology, along with evaluation of the oesophagus using barium swallow or endoscopy, are needed to diagnose the severity of the case and plan appropriate treatment. Treatment with iron supplementation can lead to the improvement of symptoms and regression of oesophageal webs, emphasising the importance of timely intervention and long-term surveillance. This case report aims to highlight the various oral and systemic manifestations, investigations, treatment, and follow-up of PVS in two female patients. The highlight of the cases is timely medical intervention leading to the disappearance of webs, eliminating the need for mechanical dilatation. Thus, early diagnosis and prompt management of such cases can improve the overall quality of life of such patients.

Keywords: Iron deficiency anaemia, Paterson-Kelly syndrome, Precancerous condition

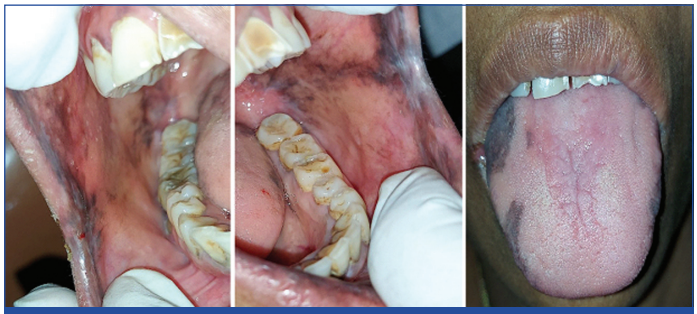
CASE REPORT

Case 1

A 53-year-old female presented with a primary complaint of fractured upper front incisors. She reported frequent fatigue and difficulty in swallowing solid food for the past three years. The patient was of slender build and presented with pale lower palpebral conjunctiva, brittle nails, and koilonychia [Table/Fig-1]. Intraoral examination revealed erosive areas with mild crusting bilaterally at the commissures of her lips, suggesting angular cheilitis. There was evidence of atrophy of the filiform papillae on the dorsal surface of the tongue and patchy brownish hyperpigmentation throughout her oral mucosa [Table/Fig-2]. Based on these findings, a provisional diagnosis of Ellis & Davey's Class II fracture of tooth 21 and anaemic stomatitis with a possibility of PVS was made. Informed consent was obtained, and the patient underwent haematological examination. It revealed a low haemoglobin level of 5.1 g/dL, and reduced values for Packed Cell Volume (PCV), Mean Corpuscular Volume (MCV), Mean Corpuscular Haemoglobin (MCH), Mean Corpuscular Haemoglobin Concentration (MCHC) [Table/Fig-3], with a peripheral blood smear showing microcytic hypochromic red blood cells. To evaluate dysphagia, a barium swallow examination was performed, which showed a single cricopharyngeal web at the C5 level [Table/Fig-4], confirming the diagnosis of PVS.



[Table/Fig-1]: Clinical images show lean built, pale palpebral conjunctiva and koilonychias.

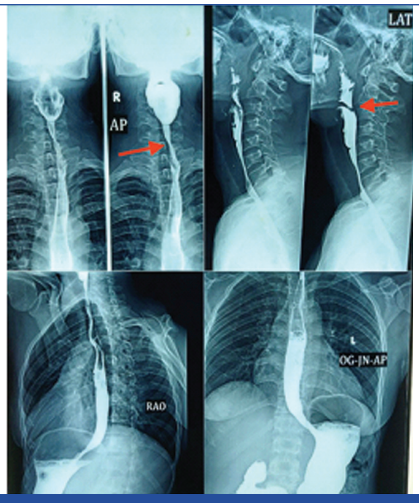


[Table/Fig-2]: Clinical images show angular cheilitis, atrophy of tongue and oral mucosa with hyperpigmentation.

Parameters	Case 1	Case 2	Reference range
Haemoglobin	5.1 gm/dL	5.1 gm/dL	12.0-15.0 gm/dL
RBC count	3.23 mill/cc mm	4.09 mill/cc mm	3.8-4.8 mill/cc mm
WBC count	7010 cells/mm^3	6400 cells/mm^3	4000-11000 cells/mm^3
Platelet count	3.94 lacs/mm^3	3.01 lacs/mm^3	1.5-4.5 lacs/mm^3
PCV	20.4%	20.1%	36-46%
MCV	63.2 FL	49.2 FL	83-101 FL
MCH	15.8 pg	12.6 pg	27-33 pg
MCHC	25.0 gm/dL	25.6 gm/dL	31.5-34.5 gm/dL

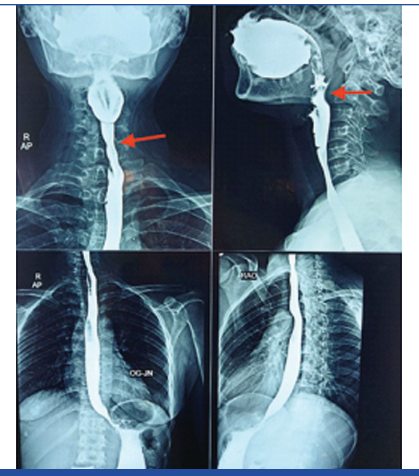
[Table/Fig-3]: Haematological examination findings of Case 1 and Case 2.

As the patient refused blood transfusion because of fear of allergic reactions and needle pricks, she was not referred to department of General Medicine and was started on therapeutic iron therapy: ferrous sulfate 300 mg once daily, folic acid 2.8 mg once weekly [1],



[Table/Fig-4]: Radiographical image shows a single cricopharyngeal web present at the C5 vertebral level in barium swallow examination.

and an iron-rich diet. After two months, she was symptomatically better, and her haemoglobin increased to 7.1 g/dL. The patient was instructed to continue the medication with monthly reviews. She adhered to treatment, and her haemoglobin level at the six-month follow-up was 12.4 g/dL. Cricopharyngeal webs were absent in the post-treatment barium swallow examination [Table/Fig-5]. The patient was advised to stop medicines and to continue her iron-rich diet. The patient subsequently underwent a composite restoration of 21 and is currently on yearly follow-up. The treatment timeline is summarised in [Table/Fig-6].



[Table/Fig-5]: Post-treatment image shows absence of Cricopharyngeal webs in barium swallow examination.

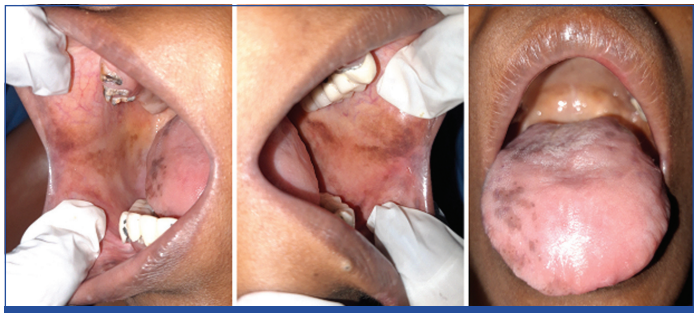
Initial Findings	At 2 months	At 6 months
Pigmentation, depapillation and dysphagia	Improvement in clinical signs and symptoms	Asymptomatic with no dysphagia
Haemoglobin-5.1g/dL	Haemoglobin-7.1g/dL	Haemoglobin -12.4g/dL
Cricopharyngeal web at C-5 level		Cricopharyngeal web absent

[Table/Fig-6]: Patient response to treatment.

Case 2

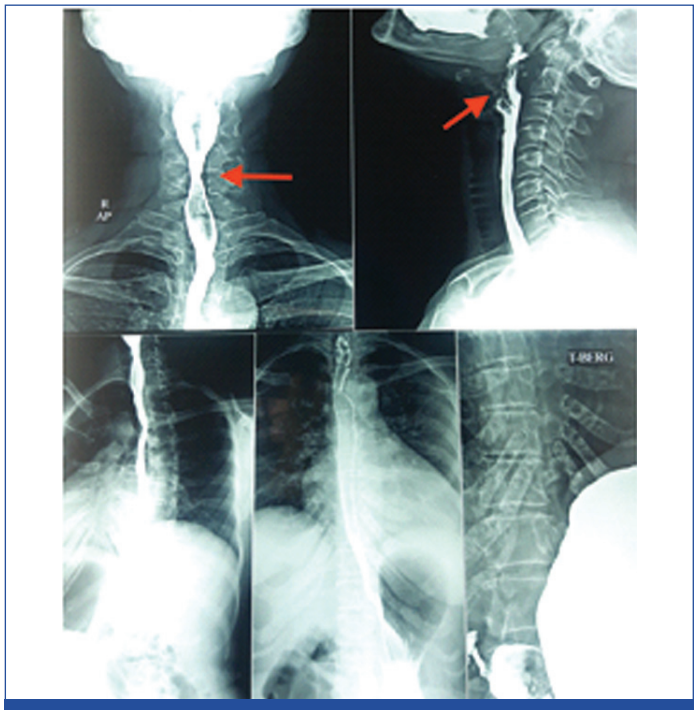
A middle-aged female presented with a complaint of a burning sensation in her mouth for the past six months and an unesthetic appearance due to a fractured prosthesis in her upper front tooth region. Her medical history revealed hypomenorrhoea. She appeared underweight and malnourished as a consequence of difficulty in swallowing large morsels. On extraoral examination, she had pallor of the palpebral conjunctiva, angular cheilitis, and koilonychia in all her fingers, implying anaemia [Table/Fig-7]. Intraoral inspection revealed pallor of the mucosa with irregular hyperpigmentation and atrophy of the papillae over the entire dorsum of the tongue [Table/Fig-8]. Based on the history and clinical examination, a provisional

[Table/Fig-7]: Clinical images show lean built, pale palpebral conjunctiva and koilonychia.



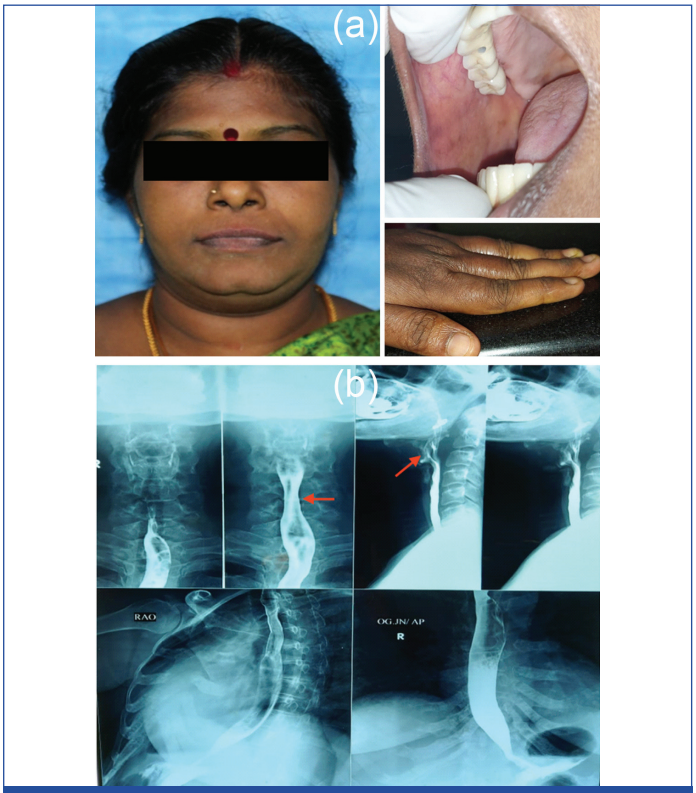
[Table/Fig-8]: Clinical images show angular cheilitis, atrophy of tongue and oral mucosa with hyperpigmentation.

diagnosis of anaemic stomatitis and fractured Fixed Partial Denture (FPD) in relation to 12,11,21 was given. PVS and Gastroesophageal Reflux Disease (GERD) were considered as differential diagnoses. A complete haemogram reported a low haemoglobin level of 5.1 g/dL and low values for PCV, MCV, MCH, and MCHC [Table/Fig-3], followed by a peripheral blood smear examination that exhibited microcytic hypochromic anaemia. Radiological examination consisted of a barium swallow test, which revealed shelf-like anterior filling in the C4-C5 level in the hypopharynx, suggestive of the presence of a small hypopharyngeal web [Table/Fig-9]. A final diagnosis of PVS was made after correlating the history, clinical manifestations, and haematological and radiological examinations. As the patient had an aversion to blood transfusion due to transfusion-transmitted infections, she was not referred to department of General Medicine and was started on iron supplementation: ferrous sulfate 300 mg once daily, folic acid 2.8 mg once weekly [1], and topical clotrimazole 1% once daily along with an iron-rich diet for one month.



[Table/Fig-9]: Radiographical image shows the presence of a shelf-like anterior filling, a small hypopharyngeal web at the C4-C5 level in barium swallow examination.

After one month, the patient was asked to repeat her haemogram, which reported an increased haemoglobin level of 7.1 g/dL. Hence, she was advised to continue the same medication for the next three months. The patient was advised to refabricate the FPD in relation to 12,11,21. At the subsequent follow-up, she appeared healthy and nourished with the absence of pallor and koilonychia [Table/Fig-10a]. By the end of three months, her haemoglobin level had increased to 11.59 g/dL, and the barium swallow test was repeated, which showed the absence of hypopharyngeal webs [Table/Fig-10b]. The patient remains healthy with periodic follow-up and a well-supplemented diet.



[Table/Fig-10]: a) Post-treatment image shows healthy and nourished, with the absence of pallor and koilonychia; b) Post-treatment image shows the absence of a small hypopharyngeal web in barium swallow examination.

DISCUSSION

The PVS, also called Paterson-Kelly syndrome, was first described in 1912. In 1939, Jan Waldenström and Sven Kjellberg described the radiological presentations of PVS [2,3]. Although the exact aetiopathogenesis of PVS is unknown, iron deficiency is the most commonly postulated aetiology, with proven improvement of webs upon iron supplementation [4]. Other theories relating to PVS include genetic factors that make individuals vulnerable to anaemia or autoimmune diseases such as coeliac disease, rheumatoid arthritis, systemic lupus erythematosus, and autoimmune gastritis [3,5].

Clinical signs of IDA include pallor of the conjunctiva and oral mucosa, brittle nails and koilonychia, glossitis, and diffuse oral mucosal hyperpigmentation. Dysphagia and glossodynia are present in cases of moderate to severe anaemia. Atkinson's classification is used to grade dysphagia based on the patient's ability to swallow food, with grade 0 indicating the ability to eat a normal diet and grade 4 depicting severe/total dysphagia [4]. The cases described above showed grade 2 dysphagia, as they could swallow semisolid foods.

Haematological investigations for IDA show low values of haemoglobin, MCV, MCH, serum iron, and ferritin, with an increased Total Iron-Binding Capacity (TIBC) [6]. Peripheral blood smear shows microcytic hypochromic red blood cells. Evaluation of oesophageal webs is commonly performed using a barium swallow examination and, in some cases, endoscopy or video fluoroscopy. The cases presented here likewise showed low haemoglobin values, with similar peripheral smear and barium swallow findings. A barium swallow examination uses barium sulphate, which provides good contrast and has been the widely used investigative modality for the detection of webs [3]. Video fluoroscopy, or video cineradiography, helps to visualise even small webs and can differentiate a true web from a false web. Upper gastrointestinal endoscopy has also been used for the detection of webs [2,4]. It is mandatory to rule out other causes of dysphagia, such as vascular rings, pharyngeal pouch and diverticulum, oesophageal strictures secondary to trauma, GERD, oesophageal motility disorders, foreign bodies, and oesophageal malignancies, before arriving at a diagnosis of oesophageal webs secondary to PVS [2]. Biopsy is recommended during endoscopy for evaluating dysplasia, as was done in a case of PVS previously reported by Mikhail D et al., who suffered from decompensated cirrhosis [7].

Treatment of PVS aims to improve the haemoglobin level, related blood parameters, and elimination of oesophageal webs [3,5]. The cases presented show a gradual increase in haemoglobin level over a period of three to six months when managed medically with iron supplements. The present report adds to the literature by describing successful reversal of anaemic signs and symptoms, including the disappearance of webs, achieved solely through comprehensive medical management in both cases without the use of any mechanical dilatation [8]. Patients who are unable to tolerate oral iron and develop epigastric distress, constipation, flatulence, or diarrhoea may be given intravenous iron therapy or blood transfusions. Oesophageal webs are generally treated with endoscopic dilatation or balloon dilatation [4], laser excision, dilatation using a cuffed endotracheal tube, and electrocautery [3,9,10]. A case of PVS previously reported by Harmouch F et al., in an octogenarian showed progression of dysphagia despite being started on iron supplementation. The patient eventually underwent Oesophagogastroduodenoscopy (EGD) to remove the food bolus stuck in her cervical oesophagus [11]. A similar case of PVS reported by Asaad A et al., in an Omani woman showed improvement in dysphagia and odynophagia post-EGD, coupled with blood transfusion and intravenous iron supplementation for two weeks [12].

PVS is grouped under Oral Potentially Malignant Disorders (OPMD), with a malignant transformation rate of 3-16%, which persists even after elimination of the webs [13]. Although PVS has a good prognosis with adequate treatment, prognosis worsens when there is an association with squamous cell carcinoma arising from the oesophageal tract [9].

CONCLUSION(S)

The PVS is a severe manifestation of IDA that can be successfully managed with timely medical intervention using iron supplements. Patient compliance with regular follow-ups and continuous monitoring is essential because of its inherent potential for malignant transformation.

Data availability: The images of investigations and clinical photographs have been uploaded and provided as part of the manuscript.

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