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Peliosis Hepatis and Iron Deficiency – An Interesting Case Report

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Authors' contributions

This work was carried out in collaboration among all authors. Authors AA and BS designed the study, wrote the protocol and wrote the first draft of the manuscript. Authors SR and VKB managed the literature searches. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Peliosis hepatis (PH) is a rare condition showing presence of multiple blood-filled cystic cavities in the liver. It does not have any gender predilection, and is suspected to be idiopathic. However, in patients with predisposing diseases, its prevalence can range from 0.2 to 22%. The association between PH and anemia has not been completely established. PH has been reported in a patients with hematologic disease, and also in patient with spherocytic haemolytic anemia. It is also suggested that acute sequestration of blood can happen in these sinusoidal cavities, which can lead to development of anemia and thrombocytopenia. We present a case of 40-year female who presented with abdominal pain and recurrent iron deficiency anemia. On examination, she was severely pale, and had moderate hepatosplenomegaly. Ultrasonography showed hepatomegaly and splenomegaly, and her blood investigations revealed severe iron deficiency with bone marrow showing hypercellular marrow with depleted iron stores. Upper gastrointestinal endoscopy was normal. Liver biopsy showed changes suggestive of peliosis hepatis. She was treated with iron and multiple blood transfusions and is in good health 6 months post presentation. Also, the association between anemia and PH has not been established.

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1. INTRODUCTION

Peliosis hepatis (PH) is a rare condition characterized by blood-filled cystic cavities, ranging between 1 mm and several centimeters in diameter [1,2]. The incidence of PH is rising due to increase in the number and quality of diagnostic methods. PH does not have any gender predilection, and is suspected to be idiopathic in nature. However, in patients with predisposing diseases such as hematologic disorders, malignancy, chronic infections (Tuberculosis, Human Immunodeficiency Virus), renal transplant, use of steroids, contraceptive pills etc, its prevalence can range from 0.2 to 22% [3].

The association between PH and anemia has not been completely established. PH has been reported in a patients with hematologic disease, and also in patient with spherocytic haemolytic anemia [4]. But, the patients were on anabolic steroids, or PH was diagnosed as an incidental finding on post-mortem examination and causality was not established. It is suggested that in patients with hematologic disorders presenting with liver disease, PH should be included in the differential diagnosis. In another report, it was suggested that acute sequestration of blood can happen in these sinusoidal cavities, which can lead to development of anemia and thrombocytopenia [5]. There is a possibility of acute bleeding into the cavities, or persistent bleed into large cavities with PH even presenting as liver hematoma [6]. However, reports regarding the causal association between PH and anemia is lacking. We report a case of PH presenting with severe recurrent iron deficiency anemia.

2. CASE REPORT

A 40-year woman presented to us with complaints of abdominal pain from last 2 years, intermittent, more in right and left hypochondrium, no aggravating or relieving factors. She also had history of easy fatigability from last 1 year. She did not have any history of blood loss, jaundice in past, pica or abdominal distention. No history of bowel or bladder disturbances. She had normal appetite, but had history of poor intake of food, and had regular menstrual cycles. Her previous investigations showed recurrent iron deficiency anemia and was treated with multiple blood transfusion and iron supplementation. No history of any other drug intake or toxin exposure. On examination, she was thin, poorly nourished with a Body Mass Index of 17.6 kg/m2. She also had severe pallor, platynychia and moderate hepato-splenomegaly. She did not have icterus, edema, lymphadenopathy or ascites. Remainder of the systemic examination was normal.

Her investigations (Table 1) revealed severe anemia with decreased reticulocyte count, microcytic hypochromic anemia and iron deficiency. Ultrasound abdomen showed presence of hepatosplenomegaly. In view of unexplained hepatomegaly, and recurrent anemia, the diagnostic algorithm planned was bone marrow biopsy, followed by upper gastrointestinal (GI) endoscopy and liver biopsy. Bone Marrow biopsy was performed in view of low reticulocyte count to rule out hypocellular marrow. However, it showed Hypercellular marrow with severely depleted iron stores. The upper GI endoscopy found to be normal, with no evidence suggestive of portal hypertension. She underwent liver biopsy to identify the etiology of hepatomegaly. It showed multiple blood-filled cystic spaces of variable sizes, present as peliotic spaces with endothelial lining, suggestive of peliosis of liver.

She was diagnosed to have PH with severe iron deficiency and was treated with 3 units of blood transfusions and iron supplementation (injectable and oral). Her hemoglobin improved to 10 g/dl, with improvement of fatigue and reduction in abdominal pain. Her etiology of the abdominal pain, which reduced with correction of anemia couldn't be exactly elucidated. After 6 months of follow up, she is stable with a Hemoglobin of 11.2 g/dl, on oral iron therapy with no further episodes of drop in hemoglobin. Her hepatosplenomegaly has also normalized at 6 months follow up. Her cause for severe iron deficiency was suspected to be nutritional deficiency, with a possibility of exaggeration by the liver lesions.

3. DISCUSSION

PH was first described in 1861 by Wagner [7] and named by Schoenlank in 1916 [8]. The etiology of PH remains unknown, but it has been reported to be associated with infectious and non-infectious causes, including drugs, chemicals, bacterial and viral infections and malignancies. Bartonella henselae is hypothesized to be the primary cause of infection [9,10]. In addition, human immunodeficiency

virus infection [11] and other wasting diseases are associated with PH. The action of vascular endothelial growth factor has been observed to be important in the pathogenesis of PH [12]. Drugs that act against PH include contraceptive steroids [13], and androgenic-anabolic steroids [14,15]. Notably, PH may present as the cardinal symptom of specific diseases, including Hodgkin's lymphoma [16]. However, the causes of PH have not be identified in 20-50% of patients [17], as observed in the current case report.

The mechanism of PH is associated with sinusoidal expansion, which is caused by obstructions in the junction of the sinusoidal and central veins of the liver. This results in focal hepatic necrosis, liver sinusoidal barrier destruction and damaged endothelial cells, as red blood cells enter the space of Disse from the sinusoids and form cystic cavities. It is difficult to differentiate other liver diseases from PH, such as abscess or carcinoma, without liver biopsy. Hence, PH should be considered in patients presenting with unexplained hepatomegaly.

We present a case of a middle-aged female with Peliosis Hepatitis presenting with recurrent iron deficiency anemia. The anemia could be an effect of PH, compounded by nutritional deficiency. However, currently there is no literature available which has studied the causal relationship between PH and anemia, as to whether anemia is the cause or effect of PH.

There are no specific treatments available for PH, however, surgery must be performed on patients with a hemorrhage, long-term medical history or limited lesions. The underlying predisposed disorders like infections, malignancy etc. should be managed accordingly. In addition, a liver transplant is necessary when patients have serious accompanying symptoms, including hepatic function failure. In these cases, the termination of any prescribed drugs is vital.

Table 1. Laboratory investigations

Test	Value	Test	Value
Hemoglobin	3.3 g/dl	Iron	13.5 ug/dl
RBC Count	2.35 million/mm3	TIBC	501 ug/dl
Reticulocyte Count	0.30%	Ferritin	2 ng/ml
Platelet count	4.32 lakhs/mm3	Vitamin B12	289 pg/ml
WBC Count	6600 cells/mm3		
PCV	12.6	RBS	109 mg/dl
Peripheral Smear	Microcytic Hypochromic anemia	Calcium	8.8 mg/dl
ESR	120 mm/hr.	Stool Occult blood	Negative
Absolute Reticulocyte count	0.10%	Stool routine	No ova/cyst
Reticulocyte Index	0.03	Urine routine & microscopy	Normal
Liver Function Tests		Total Protein	5.0 g/dl
Total Bilirubin	0.9 mg/dl	Serum Albumin	2.2 g/dl
Direct Bilirubin	0.3 mg/dl	Globulin	2.8 g/dl
AST	20 IU/L	INR	1.1
ALT	17 IU/L		
ALP	60 IU/L	HIV	Non-Reactive
		HBsAg	Negative
Blood urea	28 mg/dl	Anti-HCV	Non-reactive
Serum Creatinine	0.9 mg/dl	Peripheral smear for Malarial parasite	Negative
LDH	180 U/l	Malaria Rapid Diagnostic test	Negative
Chest X-ray	Normal		
USG Abdomen	Hepatomegaly (20cm) and splenomegaly (16cm). No evidence of dilated portal vein or hepatic biliary radicles.		

4. CONCLUSION

Peliosis Hepatis is a rare cause of liver disease and must be excluded in any patient with unexplained hepatomegaly. It is a rare condition with no specific therapy and needs close monitoring. Association between Peliosis Hepatis and iron deficiency could be causal or co-incidental and needs further evaluation.

CONSENT AND ETHICAL APPROVAL

As per university standard guideline, participant consent and ethical approval have been collected and preserved by the authors

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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