Co-Existence of Nodular Purpura and Vaccine Induced Panniculitis in a 4-Month-Old Baby-A Diagnostic Dilemma

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Abstract

Vitamin K deficient bleeding (VKDB) disorder is a rare but fatal disorder that needs prompt diagnosis and timely intervention. Among its varied clinical manifestations, nodular purpura is rare one. Proper knowledge of this presentation helps clinicians to exclude other close differentials and avoid unnecessary delays in diagnosis. Though bleeding from the vaccination site could be a manifestation of VKDB, the concomitant presence of nodular purpura and vaccine-induced panniculitis in the same patient is rare. We report a case of a 4-month-old baby presenting with both, posing a diagnostic dilemma.

Keywords: *Nodular purpura, vaccine-induced panniculitis, Vitamin K*

Introduction

VKDB is an uncommon but potentially serious condition. Vitamin K deficiency leads to impaired production of vitamin K-dependent clotting factors namely prothrombin, factor X, IX, and VII. After the introduction of intramuscular vitamin K prophylaxis at birth the incidence and prevalence of VKDB have reduced to a great extent.[1] Skin manifestation of VKDB disorder is seen in 10 to 30 % of cases and varies from purpura, ecchymoses, bleeding from the umbilicus, circumcision site, or vaccination site.[2] We describe a 4-month-old baby who presented with deep-seated subcutaneous bleeding (nodular purpura) which is a rare presentation of this condition.[3]

Case Report

A 4-month-old male child was brought with multiple bluish nodules of different sizes over the whole body with yellowish discoloration of eyes and urine for the last 2 weeks. His parents first noticed the nodules after the child received the 3rd dose of the pentavalent vaccine. The injection site nodule was erythematous, tender, warm, firm to hard, and larger than other nodules measuring approximately 2 cm in diameter [Figure 1]. The nodules at other body parts were of different sizes, non-tender, non-compressible, firm, and bluish-black in color [Figure 2]. General examination revealed mild pallor and icterus. Systemic examination revealed hepatosplenomegaly.

The child was born out of non-consanguineous marriage, a term baby with a birth weight of 2.5 kg. The baby was delivered at home. Proper information about vitamin K prophylaxis could not

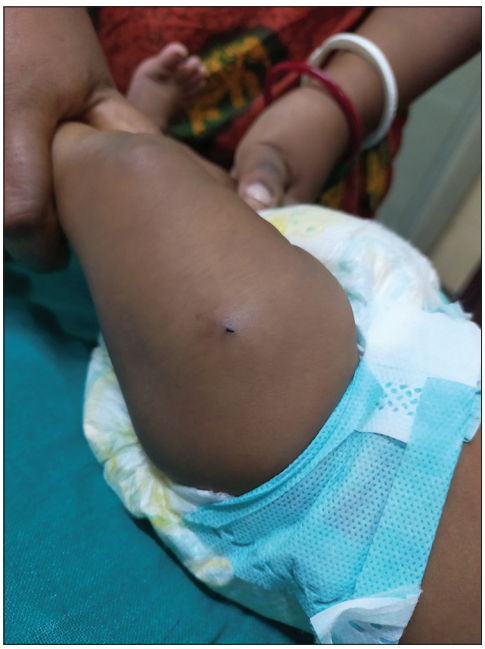

Figure 1: Erythematous firm to hard tender nodule of 2 cm diameter at the injection site of pentavalent vaccine

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be obtained. There was no history of any miscarriage or chronic medication of the mother, sibling loss, or any family history of bleeding disorder in both the paternal and maternal sides of the baby. The baby was exclusively breastfed and his vaccination status was up to date as per the national immunization schedule (NIS). The developmental milestones were as per his age. There was no history of fever, lethargy, poor sucking, vomiting, or diarrhea.

Based on the clinical findings we thought of a few differentials viz, late-onset VKDB disorder with cholestasis, hemophilia, idiopathic thrombocytopenic purpura (ITP), sepsis, blue rubber bleb nevus syndrome, blueberry muffin lesion, etc. We took an incisional biopsy from the vaccination site lesion and sent it for histopathology.

Complete blood count (CBC), peripheral blood smear, and C-reactive protein were within normal limits. Total bilirubin was 8.86 mg/dl (normal up to 1 mg/dl) with a conjugated fraction being 6.76 mg/dl (normal range: 0.00-0.2 mg/dl), and unconjugated fraction being 2.1 mg/dl (normal range: 0.1-1 mg/dl). Alkaline phosphatase was 503 U/L (<449 up to 6 months of age). The coagulation profile revealed raised PT (prothrombin time)-18.1, APTT (activated partial thromboplastin time)-62, and INR (international normalized ratio)-1.5, though fibrinogen and fibrinogen degradation product levels were normal. Direct Coomb's test and TORCH (toxoplasmosis, others (syphilis, hepatitis B), rubella, cytomegalovirus, herpes simplex) screening for congenital infections were negative. Blood and urine cultures were sterile. Urine and stool for routine and microscopy were normal. Chest X-ray (PA view), upper gastrointestinal tract endoscopy, colour doppler, ultrasonography (USG) of skin lesion and axial X-ray, X-ray of hands, feet, and long bones were normal. USG abdomen revealed cholelithiasis and hepato-splenomegaly. Histopathology of the vaccination site lesion revealed septo-lobular panniculitis with a dense inflammatory cell infiltrate [Figures 3 and 4]. The septae were thickened by fibrosis and there was lipo-membranous change and areas of hemorrhage in the subcutaneous fat. The inflammatory cell infiltrate comprised an admixture of histiocytes, lymphocytes, and plasma cells [Figure 5].

Based on the complete workup of the case, we made a diagnosis of late-onset VKDB disorder with obstructive pathology in the liver and panniculitis at the vaccination site. We administered the child three doses of intramuscular vitamin K injection at a dose of 0.5 mg/kg/day. The patient improved dramatically within 12 hours of the first dose with the disappearance of all skin lesions except the vaccination site lesion. Followed up with the patient for the next 6 months during which there was no recurrence of haemorrhage. There was the gradual resolution of panniculitis without any active intervention.

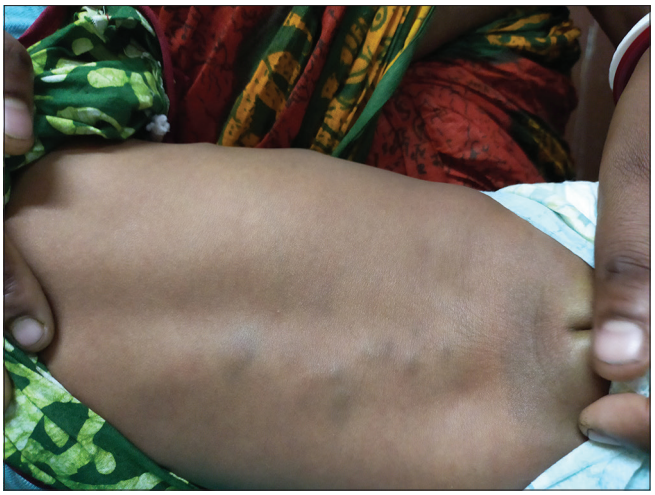

Figure 2: Variable sized bluish black nodules over the back of the baby

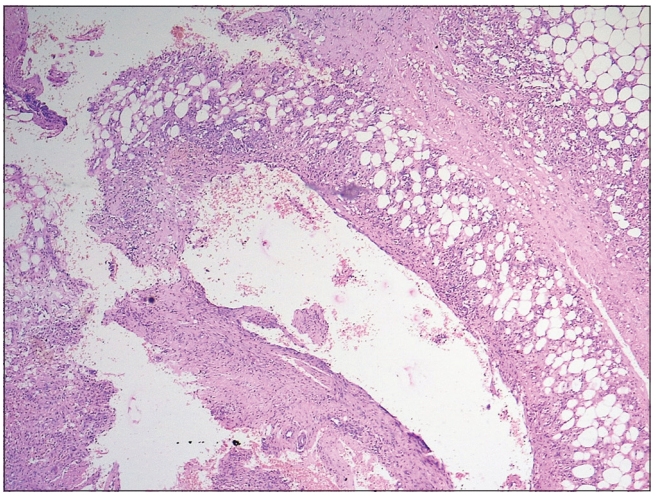

Figure 3: Septolobular panniculitis with a dense inflammatory cell infiltrate. The septae are thickened by fibrosis and there is lipo-membranous change and areas of hemorrhage in the subcutaneous fat (H&E, 100x)

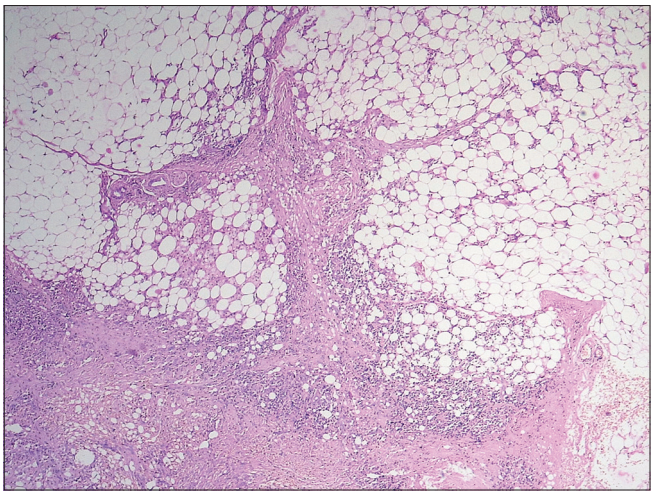

Figure 4: Septolobular panniculitis with a dense inflammatory cell infiltrate. The septae are thickened by fibrosis and there is lipo-membranous change and areas of hemorrhage in the subcutaneous fat (H&E, 100x)

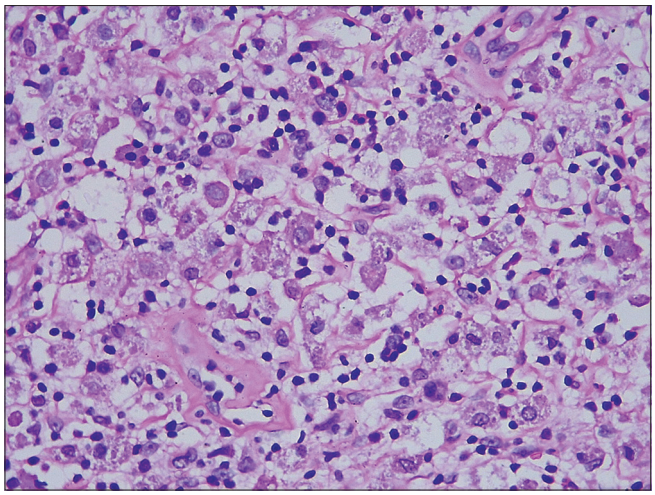

Figure 5: The inflammatory cell infiltrate comprises of an admixture of histiocytes, lymphocytes and plasma cells (H&E, 400x)

Discussion

Maternal breast milk is usually deficient in vitamin K. Late onset VKDB disorder (onset beyond seven days of age to six/twelve months of age) can occur due to exclusive breastfeeding or secondary to malabsorption (diarrhea, cystic fibrosis) and decreased synthesis or excretion (cholestasis, viral hepatitis). In our case, needle trauma acted as a triggering factor. The pathology started with exclusive breastfeeding and intensified with liver obstructive pathology.[4] Vitamin K prophylaxis may have been missed due to home birth which could have started the deficient state in the first place. Nodular purpura as a presenting feature in VKDB is rare and can cause diagnostic confusion with a list of differentials stated before. Elevated PT and APTT, absolutely normal CBC including platelets, normal peripheral smear, and dramatic response to high dose vitamin K favored our diagnosis.

Reports of vaccine-induced panniculitis are not plenty. After an extensive literature search, we were able to find few reports with cancer immunotherapy[5] and the H1N1 vaccine.[6] Evidence was strongest for the occurrence of erythema nodosum with a number of vaccines such as hepatitis B, rabies, pneumococcal, and typhoid.[7,8] Components like vaccine antigens or vehicles have been alleged to mounting abnormal sustained immune responses leading to chronic inflammation. Our case also points towards similar pathomechanism. Panniculitis following the pentavalent vaccine is rare. Proper awareness about the unusual presentation of nodular purpura is essential to exclude differentials and avoid unnecessary interventions.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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