Perianal bulla: An unusual presenting finding of necrotizing enterocolitis

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Sir,

Necrotizing enterocolitis (NEC) is the most common surgical emergency in neonates. Although clinical findings of NEC vary with the duration and extent of the disease, perianal bulla is an unusual presenting finding for clinicians. In this brief correspondence, we present the first case of NEC presented with perianal bulla. A male infant was born via cesarean delivery at 36 weeks of gestation with a birth weight of 2240 g to a 36-year-old mother who developed preeclampsia during her pregnancy. Enteral feeding was started with formula milk. The patient was consulted to our department with findings of abdominal distention, vomiting and bloody stools at 2 days of age. Abdominal roentgenogram revealed dilated loops of intestine and edema of the intestinal wall. His condition was diagnosed as a suspected NEC, and enteral feeding was discontinued with adding broad spectrum antibiotics. At 7 days of age, clinical deterioration was determined. Physical examination revealed perianal bullae [Figure 1a], and abdominal roentgenogram showed extensive linear pneumatosis of colon and ileum, in contrast to previous radiographic findings [Figure 1b]. Unfortunately, the patient died in spite of all our efforts within a few weeks.

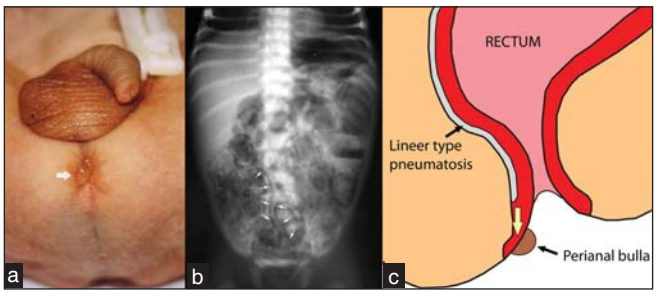


Figure 1: (a) A photo showing perianal bullae. (b) Abdominal roentgenogram showing linear type pneumatosis intestinalis (white arrows). (c) Air dissects along the rectum wall, and reaches the perianal region presenting as perianal bulla

NEC represents a disease spectrum presenting with both intestinal and systemic findings. It is widely accepted as a complication of prematurity. It is also identified that infectious agents and formula feeding play a role in the pathogenesis of NEC.[1] However, Bashiri *et al*.[2] found maternal hypertensive disorder to be risk factor for NEC. In this respect, maternal preeclampsia may be a contributing factor for development of NEC as well as prematurity and formula feeding in our case. The diagnosis of NEC is based on clinical findings as well as characteristic radiologic findings. Abdominal distention, vomiting and bloody stools are common clinical symptoms as we observed in our case. However, perianal bulla is an unusual presenting finding of NEC. Air-fluid levels, dilated loops of intestine and edema of the intestinal wall constitute common radiographic findings.[1] Despite these non-specific findings, pneumatosis intestinalis, which represents gas within the wall of gastrointestinal tract, is highly suggestive finding for NEC. There are two types of pneumatosis: Cystic (submucosal) or linear (subserosal). Several etiologies have been suggested for pneumatosis intestinalis. The formation of pneumocysts results from the interaction of multiple factors, such as mucosal integrity, intraluminal pressure, bacterial flora and intraluminal gas.[3,4] Bowen[5] propounded a causal relationship between NEC and anal fissure, and proposed that fissures might have been secondary manifestations of enterocolitis, as pneumatosis involved the rectum. We determined extensive linear type pneumatosis of colon in the present case. In this respect, we speculated that, air within the subserosal plane dissects along the rectum wall and reaches the perianal region presenting as perianal bulla [Figure 1c].

As a conclusion, perianal bulla is an unusual presenting finding of NEC. To the best of our knowledge, this is the first report suggesting an association between NEC and perianal bulla. However, retrospective and prospective studies are needed to confirm this association.

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