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Case reports

Neonatal lingual and gastrointestinal mucormycosis in a case of low anorectal malformation—a rare presentation

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Abstract We report a 1.7 kg male infant with a low anorectal malformation treated at an outside facility and referred to us on postoperative day 11. At presentation, his upper abdomen was distended, and he had perianal mucoid discharge. The tongue had a blackish discoloration. An erect abdominal radiograph showed a few fluid-filled bowel loops in the upper abdomen with a gasless lower abdomen and pelvis, suggestive of upper small bowel obstruction. There were no specific radiological features of necrotizing enterocolitis. He underwent laparotomy and bowel resection for perforated jejunum. Histopathology of the tissue specimen was suggestive of mucormycosis. Postoperatively, he received intravenous amphotericin B (liposomal) and was started on liquid enteral nutrition after 2 weeks. However, the anterior two thirds of his tongue gradually sloughed off. He is awaiting reconstruction of the tongue. The purpose of this report is to emphasize that physicians should have a high index of suspicion for oral and gastrointestinal tract mucormycosis in neonates with metabolic disturbances who present with a discolored oral mucosa and an abdominal mass with intestinal obstruction. Early diagnosis and an aggressive approach of combined medical and surgical treatment may improve the outcome of patients with this potentially lethal invasive disease.

Mucormycosis is an infection caused by fungi belonging to the order Mucorales of the class Zygomycetes. They are ubiquitous organisms found in the soil and decaying organic matter. The genera causing disease in humans include *Mucor*, *Rhizopus*, and *Absidia* [1]. These fungi produce airborne spores that are the infective form. Infections occur in the rhinocerebral, respiratory, gastrointestinal, or cutaneous regions, depending on whether the spores are inhaled, ingested, or injected.

Mucormycosis infection is seen in conditions associated with compromised immunity in adults with diabetic ketoacidosis, lymphomas, leukemias, and renal failure on peritoneal dialysis [2]. The predisposing conditions in children are prematurity, neutropenia, acidosis, and corticosteroid therapy.

Mucormycosis of the gut in neonates is rare, difficult to recognize, and, hence, usually fatal. These cases are usually diagnosed initially as necrotizing enterocolitis (NEC), resulting in delay in specific treatment. The presence of mucormycosis at rare sites, such as the tongue, can be easily missed by a physician who is unaware of such a presentation. We report a case of lingual and neonatal gastrointestinal tract (GIT) mucormycosis diagnosed after laparotomy, with a

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review of cases from the literature of lingual and GIT mucormycosis in newborn infants.

1. Case report

A 1.7 kg male infant was born to a primigravida mother at 41 weeks of gestation by normal abdominal delivery. There was no significant antenatal history. The infant developed abdominal distension and bilious vomiting soon after birth and did not pass meconeum. He was admitted to a neonatal intensive care unit at an outside facility and was diagnosed with a low anorectal malformation. A cut back anoplasty was performed. He was discharged on the second postoperative day. He was asymptomatic, feeding well on breast milk, and passing stools regularly. To compensate for the poor sucking, the child was fed with expressed breast milk (collected in a small bowl) with a spoon for 2 to 3 days before presenting to us. The infant's weight was low because the mother was anemic and malnourished. The mother was illiterate and lived in a low socioeconomic environment. On postoperative day 11, he developed bilious vomiting and abdominal distension and was readmitted. He deteriorated rapidly with the onset of metabolic acidosis, hyponatremia, and thrombocytopenia. His platelet count was 40,000 mm^3, and C-reactive protein level was 76. He was referred to us with suspicion of intestinal obstruction. On physical examination, his upper abdomen was distended, and he had perianal mucoid discharge. His tongue had a blackish discoloration (Fig. 1A). At presentation, the infant was severely dehydrated, and his arterial blood gas analysis was suggestive of severe metabolic acidosis. A nasogastric tube was inserted and placed on suction, and he was started on parenteral fluids and antibiotics. He received blood, platelets,

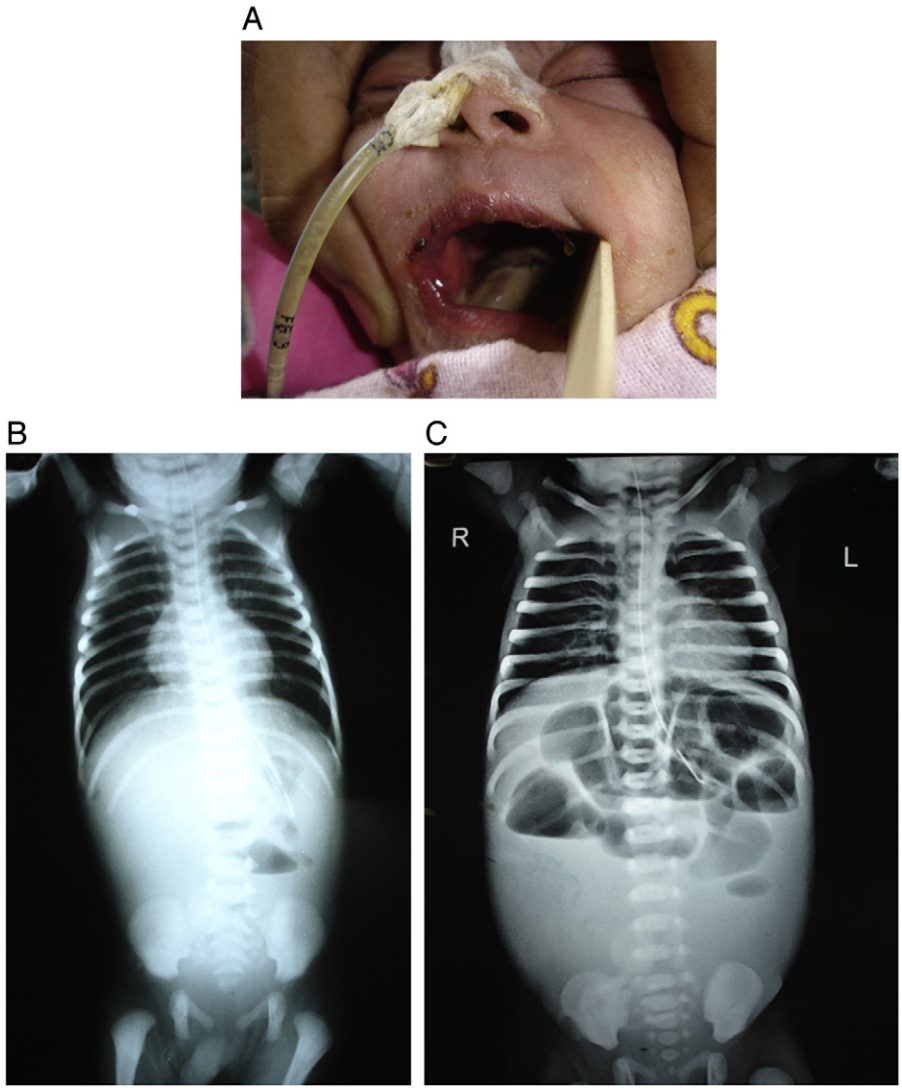

Fig. 1 A, Tongue showing blackish discoloration. B, Erect abdominal radiograph (at admission) showed a few bowel loops in the upper abdomen with gasless lower abdomen and pelvis, suggestive of upper small bowel obstruction. C, Follow-up erect abdominal radiograph on day 2 of admission showed multiple fluid-filled bowel loops with a gasless pelvis.