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Postoperative Intestinal Perforation Secondary to Mucormycosis Following Ventricular Septal Defect Closure: A Rare Complication

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Abstract

Intestinal perforation is a rare complication after Ventricular Septal Defect (VSD) closure. Mucor mycosis is a rare opportunistic infection, usually found in immunocompromised patients. It is a primitive fungus which is saprophytic & ubiquitous with a fast growth rate. In humans it shows a rapid evolving course, tissue necrosis & blood vessel invasion. We report a case of intestinal perforation as result of gastrointestinal Mucor mycosis infection during post-operative period of VSD closure in a non-immunocompromised infant.

Keywords

Mucormycosis, Ventricular Septal Defect, Peritoneal Dialysis, Jejunostomy, Health Care Associated Infection

1. Introduction

Mucormycosis enters the human body via upper respiratory tract, oral cavity, hematogenous spread, through contaminated surface wounds and also through IntraVenous access sites. Gastrointestinal tract is the most common site of involvement in preterm neonates, causing necrotizing enterocolitis and intestinal perforation by invasion of underlying bowel vasculature, whereas in the older children and adults it affects lungs, nasal sinuses and skin [1] [2]. We present a case of a postoperative VSD closure child with bowel infarction and multiple intestinal

perforations due to intestinal Mucormycosis infection.

2. Case Summary

A 3 months old male child diagnosed with Acyanotic Congenital Heart Disease (ACHD) VSD, Atrial Septal Defect (ASD) and Patent Ductus Arteriosus (PDA) at 15days of life, presented to the emergency department with complaints of fast breathing and shallow breathing immediately after spoon feeding. On examination, child had tachycardia, tachypnea and was irritable with intercostal and subcostal retractions, bilateral equal air entry with crepitations more in the left lung fields compared to the right side. Cardiovascular examination revealed hyperdynamic precordium, with pan systolic murmur lateral to left lower sternal border with loud Pulmonary component of second heart sound and fixed wide split with small hemangioma of 1 × 1.5 cm in the epigastric region. Child was clinically diagnosed as a case of ACHD with aspiration pneumonia.

2-Dimensional Echocardiography demonstrated large malaligned perimembranous VSD with inlet extension of 1 × 1 cm size, ASD with left-to-right shunt, moderate to severe Pulmonary Artery Hypertension, dilated Right Atrium and Right Ventricle and normal biventricular function.

Child was managed initially with non-invasive positive pressure ventilation, feeds given via Naso Gastric tube and broad-spectrum intravenous antibiotics, Diuretics, Angiotensin Converting Enzyme inhibitors and Proton Pump Inhibitors. Child improved symptomatically and ventilatory support was weaned off. Fluoroscopy was done to rule out Tracheo Esophageal Fistula. Elective closure of VSD via Trans Right Atrial route along with ASD closure and Ductus ligation, was performed and small Patent Foramen Ovale was left in situ. Child was shifted to Intensive Care unit (ICU) on inotropic support of Dobutamine @ 6 mcg/kg/min, Adrenaline @ 0.05 mcg/kg/min and Nitroglycerine @ 0.8 mcg/kg/min and elective invasive mechanical ventilation. On postoperative day 1, child developed Low Cardiac Output Syndrome (LCOS) manifested by hypotension, elevated lactate levels, decreased urine output and increased core temperature. Peritoneal dialysis was started and gradually LCOS improved and inotropes and mechanical ventilation were weaned off. Peritoneal dialysis catheter was removed on 6th postoperative day.

On postoperative day 7, child developed abdominal distention and feculent output from the peritoneal dialysis catheter insertion site. Child was transferred under the care of pediatric surgery and exploratory laparotomy was performed. Intraoperative findings were malrotation of mid gut and intestinal perforation at distal ileum for which ileal resection, distal ileostomy and Ladd's procedure were done. On pod 5 after an exploratory laparotomy child again developed fecal discharge from drain site and exploratory laparotomy was performed for the second time. Intraoperative findings were: two spontaneous intestinal perforations in jejunum for which Jejunal resection and Jejunostomy were performed (**Figure 1**). Biopsy of the resected intestine revealed intestinal Mucormycosis, and intrave-

nous Liposomal Amphotericin-B was administered @ 5 mg/Kg/day slowly over 2 hours for total duration of 6 weeks. Jejunostomy being a high output stoma, child was managed with Total Parenteral Nutrition. After gaining a minimal adequate weight from 3.6 to 3.8 kg, jejunostomy closure was performed. On POD 3 of Jejunostomy closure, Ileostomy stoma started functioning and on POD 5 sips of clear liquids were allowed, child tolerated oral feeds well and feeds were increased gradually. Child's nutritional status improved and feeding plan was devised as per dietician and pediatrician's advice. Semisolid diet was allowed and child gradually gained weight and stoma output was formed solid stool. After 5 months of hospital stay, child was discharged with ileostomy and elective ileostomy closure was performed after 3 months, which was uneventful.

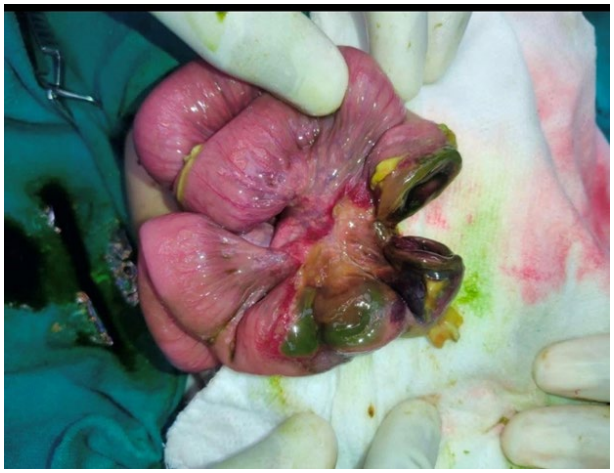


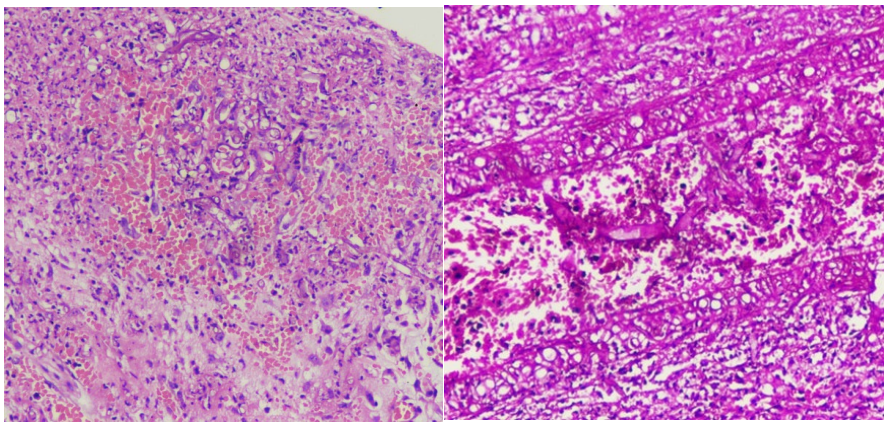
Figure 1. Resected gross specimen of small intestine with multiple perforations.

3. Discussion

Higher incidence of Mucormycosis is seen in immunocompromised patients those with diabetes mellitus (40%), hematologic malignancy (33%), post organ transplantation (approx. 25%), corticosteroid therapy, neutropenia, recent surgical history, trauma (major and minor) and in burns patients [1].

In the gastrointestinal tract stomach is the most common site of mucormycosis infection comprised of 67%. Colon is the second most common site of involvement comprising of 21%. Involvement of small intestine and esophagus is rarely seen [3]. Clinical presentation of Gastrointestinal Mucormycosis is very aggressive with and a high index of suspicion is needed. Gut Mucormycosis infection is suspected in preterm and low birthweight babies with clinical manifestations similar to that of necrotizing enterocolitis in the form of rapid onset abdominal distention, not tolerating feeds, low serum sodium levels, elevated White Blood Cell (WBC) count and low platelet count. But gut Mucormycosis does not have features of pneumatosis intestinalis in abdominal radiographs and does not respond to antibacterial therapy [4]. So far there is no literature available suggesting that

the diagnosis of gastrointestinal Mucormycosis has been made on clinical signs and symptoms alone. Isolating the organism by growing on culture media is not very sensitive. The gold standard diagnostic test is Histopathological examination of resected specimen for fungal hyphae using Hematoxilin and Eosin (H & E) stain (**Figure 2(a)**) and special stains such as Periodic acid-Schiff (PAS) stain (**Figure 2(b)**) and Grocott Methenamine Silver (GMS) stain (**Figure 2(c)**).



(a) (b)

Figure 2. Histology images of the resected specimen. (a) Hematoxilin and Eosin stain 10× magnification; (b) Periodic acid-Schiff (PAS) stain 4× magnification.

Intestinal mucormycosis infection detected in our case can be attributed the term "Health Care-associated" noticed in immune competent subset following prolonged hospitalization or complex surgical procedure [5] which can be attributed to presence of too many invasive lines predominantly peritoneal dialysis catheter in this case. Kesckes *et al.* opines that there are high chances for spores of Mucoraceae to grow in premature infants when they are placed in incubators with high humidity [6]. Mortality rate of gastro intestinal mucormycosis is very high as a result of bowel perforation which is about 85% [7] as a result of which diagnosis of this infection in the gastrointestinal tract is a postmortem finding in almost all the cases as reported by Chakrabarti A. *et al.* [8] and our case is a contrary to this fact.

In the present case, presence of midgut malrotation diagnosed intraoperatively could be a predisposing factor for intestinal ischemia, but multiple site perforations are unusual which can be attributed to the invasive hyphae of the fungi clogging the intestinal vasculature causing thrombosis and tissue necrosis. Michalak DM *et al.* reported a survived case of gastrointestinal mucormycosis infection which presented as perforation managed by surgical resection and intravenous Amphotericin B in a preterm neonate who was treated for Acute Respiratory Distress of newborn. Role of Amphotericin B in the treatment of gastrointestinal Mucormycosis is of debate, and it seems rationale that resection of involved bowel segment is not only a treatment to manage intestinal perforation but also eliminates the fungal infection load [9].

This kind of aggressive treatment strategy was followed even in our case and might be attributed to the survival outcome in our patient. Poyuran R *et al.* reported a case of postoperative Tetralogy of Fallot intra-cardiac repair who developed Ileo-cecal Mucormycosis induced bowel perforation managed in a similar fashion but unfortunately the patient did not survive [10].

4. Conclusion

It is understood that a high index of suspicion is required for a prompt diagnosis of this rare causative organism of bowel perforation especially in prolonged hospitalization and ICU stay cases and in postoperative patients who underwent major surgical procedures such as cardiac surgeries even though they don't have any predisposing factors to place them into the immune compromised subset of patients. As a result of which gastrointestinal Mucormycosis can be considered as a hospital acquired or nosocomial or health care associated infection.

Ethics Approval

The study has been conducted after approval from Institutional Ethics Committee of All India Institute of Medical Sciences, Raipur, India for reviewing the medical records of the patients bearing approval number IECSG-189/08-08-2023. The patient data and other information were used in compliance to the Declaration of Helsinki.

Informed Consent

Informed Consent has been obtained from the patient(s) to publish their information anonymously for academic purposes.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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