This case highlights endoscopic management of IOP secondary to malrotation with partial volvulus during pregnancy with successful outcome. This is the first case in published literature where pregnancy with malrotation and a volvulus was treated successfully non-surgically using a nasoenteric tube for feeding cum stenting.

Early establishment of definitive diagnosis even if it involves radiation exposure is key in organizing management of such problems during pregnancy. Risk of maternal and fetal mortality associated with intestinal obstruction outweighs the potential risk of fetal radiation exposure. Fetal risk is considered to be negligible at exposures of 5 rad or less when compared to the risks of intestinal obstruction during pregnancy. The radiation exposure with plain X-ray is only 0.1 rad and with CT scan it is 2.6 rad both of which are within the safe limits of radiation exposure during pregnancy.[11]

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Hirschsprung's disease in adults presenting as sigmoid volvulus: a report of three cases

Introduction

Hirschsprung's disease (HD), being congenital in origin is usually seen in infancy and early childhood. However, less commonly it is recognized in adults also.[1] Volvulus of colon is a rare emergency presentation of HD in children.[2] Adult HD presenting as sigmoid volvulus is still less common, with very few cases reported in literature.[3,4] We report 3 cases of young adults presenting with sigmoid volvulus, who on subsequent evaluation were found to have HD and offered appropriate definitive treatment.

Case 1

A 33 year old mentally subnormal male presented with history suggestive of acute large bowel obstruction. He had chronic constipation requiring frequent enemas and laxatives. Clinical and radiological evaluation was consistent with a diagnosis of sigmoid volvulus. Sigmoidectomy with colorectal anastomosis and defunctioning transverse colostomy was done. Three months later, colostomy closure was done. About one week following surgery, patient again developed symptoms of intestinal obstruction, not relieved by conservative management. With a provisional diagnosis of post-op adhesive obstruction, he was again taken up for surgery. Intra-operatively the entire large intestine was found to be enormously dilated with a perforation of size 1 cm in the descending colon. Thorough lavage of the peritoneal cavity and simple closure of the perforation was done and transverse colostomy redone. Rectal biopsy was taken which showed features of HD (**Figures 1 & 2**). Definitive surgery was planned,

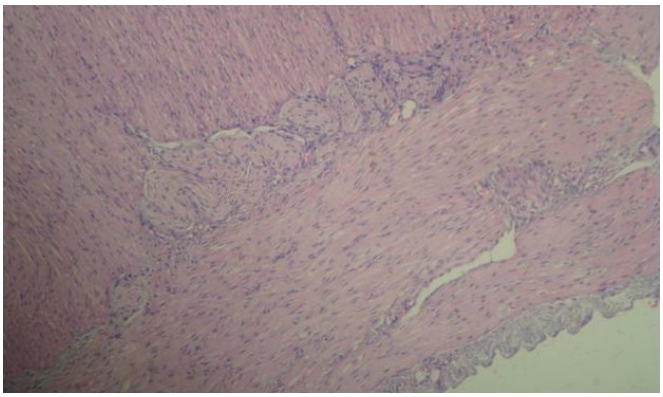

Figure 1: Photomicrograph of the rectal biopsy showing hypertrophied nerve bundles and absent ganglion cells (H&E ×10)

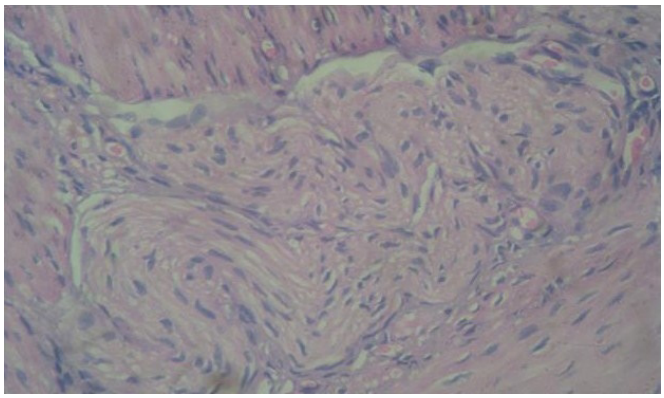

Figure 2: Photomicrograph of the rectal biopsy showing hypertrophied nerve bundles (H&E ×40)

but unfortunately, the patient expired due to septicemia caused by fecal peritonitis

Case 2

An 18 year old female presented with clinical and radiological features suggestive of sigmoid volvulus. Intra-operatively the diagnosis was confirmed. Based on our experience with the previous case and the patient being young, HD was suspected and it was decided to defer definitive surgery for volvulus until the diagnosis of HD was ruled out. Though there were no features suggestive of gangrene, sigmoid resection with double barrel colostomy was done. Rectal biopsy confirmed our suspicion of HD. Swenson's procedure was done and the patient is on follow up.

Case 3

A 24 year old male presented with features of sigmoid volvulus. Emergency decompression was done with flatus tube. Subsequent rectal biopsy showed absence of ganglion cells and hypertrophied nerve fibres suggestive of HD. Modified Soave's procedure was done as the definitive procedure and the patient is on follow up.

Discussion

Hirschsprung's disease or congenital aganglionic megacolon was first described by Harald Hirschsprung in 1886.[5] The incidence is about 1 in 5000 live births and it is characterized by total absence of intramural ganglion cells in the Meissner (submucosal) and Auerbach (myenteric) plexuses resulting from defective craniocaudal migration of ganglion cell precursors to the hindgut. In the majority (80%) of patients it is confined to the recto sigmoid region (short segment type), however occasionally it may extend to the whole colon or may be limited to a very small segment of rectum (ultra-short segment).[6] Due to its congenital origin, majority of the described cases are in newborns and infants. Delay in passage of meconium, obstipation, abdominal distension, bilious vomiting and failure to thrive are the usual presenting symptoms of HD. However milder cases do not present until adolescence or adulthood.[1] Though adult onset HD was thought to be rare, Hirschsprung himself had suggested as early as 1900, that the disease was not confined to children and several cases of adult HD have been reported since then.[7] Adults with HD usually present with life-long constipation requiring frequent laxatives and enemas.

The emergency complications of HD in children mainly include enterocolitis and obstruction; colonic volvulus being a rare complication. The first case of pediatric HD presenting as sigmoid volvulus was reported as early as 1952 by Dean and Murry.[8] Since then cases of pediatric HD presenting as colonic volvulus involving both the sigmoid[2,9,10] and transverse colon[11] have been reported.

The described complications of adult HD include severe fecal impaction leading to obstruction, superficial inflammation and ulceration of mucosa, hemorrhage, ischemia secondary to compromise of vascularity by colonic distension, perforation and volvulus of the colon secondary to an elongated colonic mesentry.[12] The first case of adult HD presenting as sigmoid volvulus was reported by Tan et al only in 2006.[3] Recently Ghaemi et al have reported four more cases.[4] We are reporting 3 more such cases. Sarioglu et al have reported a 0.66% incidence of colonic volvulus among children with HD.[9] No such reports are available in adults. An aganglionic segment

below the sigmoid colon and a freely mobile mesosigmoid seem to be responsible for the volvulus.[9]

The diagnosis of HD is usually much more difficult in adults than in infants, because of the rarity of the disease and the higher incidence of short or ultra-short segment aganglionosis in adults. Ultimately, rectal biopsy is required for making a definitive diagnosis.[1]

The standard surgical procedures for HD include the abdominoperineal pull-through described by Swenson and Bill, retrorectal pull-through described by Duhamel, endorectal pull-through described by Soave and the posterior anorectal myectomy described by Lynn.[1] Though a definitive assessment of the long-term functional outcome of the principal procedures has remained inconclusive, the Duhamel procedure has been found to be associated with lower postoperative morbidity rate, better functional outcome and wider acceptance in adult HD.[1]

Sigmoid volvulus is generally described as an illness occurring in elderly individuals, particularly those with psychiatric disorders and those residing in nursing homes and mental institutions.[13] However it can also occur in younger population. The definitive treatment for sigmoid volvulus is grossly different from that of HD and failure to recognize HD as the primary cause in a patient presenting with sigmoid volvulus would result in inappropriate and inadequate treatment. Hence it is of paramount importance that whenever a young adult presents with sigmoid volvulus, rectal biopsy must be done to rule out HD.

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Dead worm in common bile duct

Introduction

Ascariasis is a common problem in tropical countries and infestation of bile duct can occur. The worm generally continues to survive in the bile duct. However, the worm can die inside the bile duct and create a foreign body. This case reports an unusual presentation of a dead worm in the common bile duct.

Case Report

A 5-year-old boy who had undergone treatment two years ago for malnutrition due to multiple worm infestations presented