



A Case Report of a Patent Omphalomesenteric Duct Presenting with Meconium Discharge from the Umbilicus

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Abstract

This report describes the case of a newborn presenting with a patent omphalomesenteric duct remnant fistula identified at birth with meconium in the umbilical cord. The infant had no antenatal care and presented to the NICU in mild respiratory distress and meconium within the umbilical cord. We instituted a brief work up and proceeded to surgery where the remnant was resected. The patient did well and was discharged soon after without complication. Here we describe the case, the work up and a review of the literature.

Keywords: Omphalomesenteric duct fistula; Vitelline duct fistula; Ileo-umbilical fistula; Persistent patent omphalomesenteric duct; Meconium discharge from umbilicus; Exomphalos minor fistula

Introduction

Umbilical anomalies are not uncommon in the infancy and reports date back since antiquity. Newborns often present with a moist umbilicus which can be associated with various abnormalities from simple granulomas to persistent urachus. When serous, feculent or bilious drainage are identified at the umbilicus it is suggestive of a patent omphalomesenteric duct with fistulous communication with the umbilicus. Symptoms of fecal drainage (“congenital umbilical anus”) and prolapse of the intestine are well known, however infrequent. The surgeon is advised to avoid resection of the umbilical “tumor”, in the fear of exposing intestinal lumen indicating a vitellointestinal remnant.

Newborns presenting with fecal drainage from the umbilicus can be quite alarming to parents. Providing counseling and reassurance that the clinical diagnosis and subsequent treatment are both straight forward and curative. The extent of the work up and timing of treatment usually depends on the age of presentation and the ultimate diagnosis. Persistent omphalomesenteric duct remnant is a less common entity and fistula to the umbilical cord at birth remains rare. Here provide a brief review of the literature and describe a case where the patient presented at birth with an omphalomesenteric remnant fistula to the umbilical cord.

Case Presentation

This report describes the case of a newborn presenting with a patent omphalomesenteric duct fistula identified at birth with meconium in the umbilical cord. A 33 week male neonate was born to a mother with a known history of drug abuse who presented in labor without antenatal care. Maternal urine drug screen was positive for amines but the specific agent could not be identified. At birth he presented in mild respiratory distress with meconium discharge within the right side of his umbilical cord (Figure 1). There were no abdominal wall abnormalities and the remainder of the physical exam, including rectal exam, were unremarkable. Differential diagnoses included intestinal perforation, meconium peritonitis, and small omphalocele with perforation and patent omphalomesenteric duct with ileal fistula into the umbilical cord. An abdominal x-ray showed air filled small intestine without air-fluid levels or any intra-abdominal calcifications. The colon could not be confirmed on plain x-rays so contrast enema (Figure 2) was performed to interrogate the colon for any anomalies. The contrast enema showed no abnormalities and demonstrated contrast entering the umbilical cord confirming our diagnosis of patent omphalomesenteric duct remnant with ileal to umbilical cord fistula. A cardiac workup, including an echocardiogram and electrocardiogram, was negative except for a small patent ductus arteriosus.

The patient was subsequently taken to the operating room and an exploratory laparotomy through a supraumbilical omega incision was performed. An intraoperative diagnosis of a patent

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Figure 1: Meconium discharge within the umbilical cord with the arrow pointing at the mucosal surface of the fistula.



Figure 2: Contrast Enema demonstrating no intestinal atresia with arrow showing contrast entering the umbilical cord.

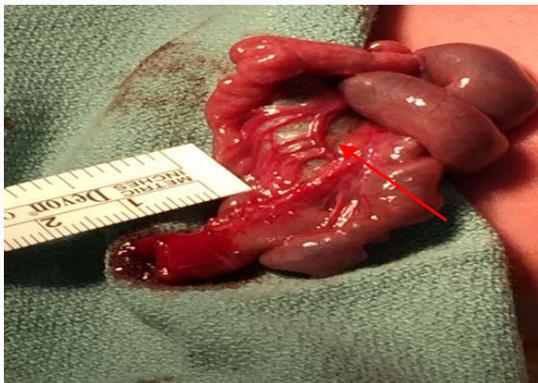


Figure 3: Intraoperative photo of the patent Omphalomesenteric duct with arrow pointing to the fistula being measured by the ruler.

omphalomesenteric duct with omphalo-ilio umbilical fistula was confirmed (Figure 3). Options for resection included a segmental small bowel resection to include the fistulous connection, a diverticulectomy across the base of the anomaly with a stapler, versus excising the base of the tract and performing a hand-sewn closure. Resection across the base of the fistula was performed by dissecting the fistulous connection away from the cord structures and stapling across the base with a JustRight 5mm stapler (JustRight Surgical, LLC, Boulder, CO, USA). This decision was purely surgeon preference to expedite and minimize the risk of stricture formation. At completion of the resection the closure was tested by milking intestinal contents across the segment of bowel under pressure. Once satisfied with the

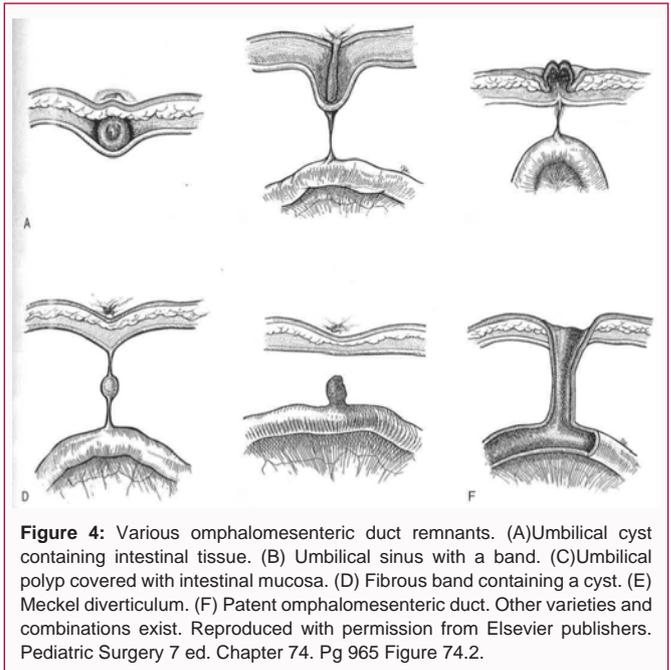


Figure 4: Various omphalomesenteric duct remnants. (A) Umbilical cyst containing intestinal tissue. (B) Umbilical sinus with a band. (C) Umbilical polyp covered with intestinal mucosa. (D) Fibrous band containing a cyst. (E) Meckel diverticulum. (F) Patent omphalomesenteric duct. Other varieties and combinations exist. Reproduced with permission from Elsevier publishers. Pediatric Surgery 7 ed. Chapter 74. Pg 965 Figure 74.2.

resection, the abdomen was irrigated and closed in the usual fashion. The defect in the umbilical skin was closed with a pursestring suture and tacked to the fascia completing the umbilicoplasty. The patient tolerated the procedure well and returned to the NICU where he began feeding and was discharged without complication.

Discussion

The Omphalomesenteric duct is a communication between the primitive midgut and the yolk sac. Normally, the duct obliterates around 6 weeks of gestation [1], yet varying degrees of incomplete obliteration can take place in 1-4% of infants [2]. Exact etiology of incomplete obliteration remains an enigma. Various teratogenic models are present in the literature however none accurately address a direct cause/effect relationship with these anomalies. In our patient the working hypothesis include maternal amine exposure throughout pregnancy which has been linked to abdominal wall defects and intestinal atresias [3].

Meckel's diverticulum, vitelline cyst, vitelline ligament, and umbilical sinus represent incomplete obliteration of the omphalomesenteric duct and are the most common forms of presentation. Figure 4 shows various forms of omphalomesenteric duct remnants with letter F illustrating our case. Meckel's diverticulum is the most common anomaly and is often asymptomatic. When symptomatic they can present with painless gastrointestinal bleeding, intestinal obstruction, intussusception and rarely Meckel's diverticulitis. An omphalomesenteric fistula, as in our patient, is a complete failure of obliteration of the omphalomesenteric duct and is a rare finding at birth. A review of the literature (Table 1) found only one case of a patent omphalomesenteric duct presenting at birth with meconium in the umbilical cord. The report by Ng J et al. [4] comprises a 10 year review which identified two cases of ileal umbilical fistula and its association with exomphalos minor. All of the other reports describe a delayed presentation and management of these umbilical anomalies. Age at presentation varied from 10 days to 8 months of age with signs and symptoms ranging from vague umbilical drainage and feeding intolerance to a red mass at the umbilicus. One could argue that patients presenting at day 10

Table 1: Case presentations of omphalomesenteric fistulas presenting in infancy.

Author	Age of presentation	History of presentation	Findings	Treatment	Outcome
Patel R et al. [6]	10 days old	Vomiting and poor weight gain and a prolapsing lesion at umbilicus	Bulky umbilical cord that showed lesion once cord separation occurred	Trans-umbilical exploration with resection of the Vitelline duct and anastomosis	Uneventful, discharged home after 48 hours. Thriving at 3 month follow-up
Konvolinka C [7]	6 weeks old	Umbilical drainage	Umbilical mass with exposed mucosa.	Laparotomy with segmental resection	Not reported
Singh S et al. [8]	8 months old	Red mass from the umbilicus for 2 days	Dusky, edematous bowel in the umbilicus.	Resection of bowel with anastomosis and repair of abdominal wall defect	Discharged on POD 9 and follow showed a healthy child.
Tamilselvan K et al. [9]	6 months old	Discharge from the umbilicus	Cyst and cutaneous fistula without communication into the bowel	Surgical resection of cyst and fistula.	Recovery was uneventful.
Giacalone G et al. [10]	10 days old	Fecal discharge from umbilicus	Protrusion of intestinal mucosa from umbilicus.	Excision of fistula and closure of the umbilicus.	No complications post-operatively and discharged home after 3 days
Ng J et al. [4]	At birth	Meconium in exomphalos minor	Meconium in umbilical sac	Small bowel resection	No complications
Maxwell D et al.	At birth	Meconium in umbilical cord	Presence of meconium in the umbilical cord with a fistula into the ileum	Stapling of the base of the fistula and resection of the fistula with umbilicoplasty.	Discharged home without complication

of life are similar to those presenting at birth. This may in fact be the case, however, we surmise that presentation at birth represent a unique subset of this population. Newborn presentation may have associated distal intestinal atresia increasing pressure across the fistula precipitating meconium discharge at birth, be variants of omphalocele, or simply represent arrest in development at an earlier stage accounting for a wider fistulous tract.

The evaluation of these patients may be as simple as making the clinical diagnosis without further investigation, may include probing the fistula tract with a blunt probe, or perhaps injecting the tract with contrast to confirm its origin. All of these patterns of investigation have been described however, we performed a contrast enema to ensure there was not a distal intestinal atresia accounting for the early presentation and the lack of distal intraluminal air on plain radiograph. We found the contrast study diagnostic and informative by demonstrating contrast exiting through the umbilical cord without evidence of distal intestinal atresia.

Although there have been rare reports of spontaneous regression of a patent omphalomesenteric duct [5], the vast majority of literature recommend early surgical resection as the appropriate course of management in order to prevent further complications from the malformation. Small bowel resection with primary anastomosis is commonly performed yet we performed a stapled resection at the base of the anomaly, previously not described, with good result.

In summary we present a case of patent omphalomesenteric duct presenting with meconium discharge into the umbilical cord at birth. Careful inspection should be performed of the base of the umbilical cord at birth to ensure the clamp isn't placed across the prolapsed mucosa of an omphalomesenteric remnant. If there is meconium within the umbilical cord then a patent omphalomesenteric duct with fistula should be considered and urgent surgical resection is indicated to prevent morbidity and possible mortality secondary to the malformation.

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