Omphalomesenteric Duct Fistula: A Rare Manifestation of Omphalomesenteric Duct Anomalies

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ABSTRACT

Omphalomesenteric duct (OMD) anomalies occur as a result of resorption failure during gestation. Various forms of OMD anomalies are documented depending upon the degree of resorption (complete or partial) including Meckel's diverticulum, fistulas, fibrous bands, and cysts to umbilical polyps. Among these, Meckel's diverticulum is the most common of these anomalies, and omphalomesenteric duct fistula (OMF) is the most rare. Complete persistence of OMD in neonates results in OMF, which has a male predominance. OMF is documented in less than 0.1% of the population. Differentials include urachal remnants and umbilical granuloma. We report a rare case of a 10-day male neonate who presented with umbilical swelling and discharge. This case is presented to highlight the occurrence of this extremely rare phenotype to raise a high index of suspicion and awareness among healthcare providers.

Key Words: *Omphalomesenteric duct fistula, Umbilical swelling, Sinogram.*

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INTRODUCTION

The omphalomesenteric duct (OMD) is a normal embryological structure between the yolk sac and primitive gut that obliterates during the 7th to 10th week of gestation. OMD anomalies represent a spectrum of entities resulting from either partial or complete lack of resorption of the omphalomesenteric duct (vitelline duct). Meckel's diverticulum is the most common manifestation of the spectrum while other less frequent anomalies include OMD fistulas, cysts, fibrous bands, and umbilical polyps, with omphalomesenteric duct fistula (OMF) being rarest.[1,2]

Vitello-intestinal duct or OMD persistence is a rare congenital malformation of the gut, with an incidence of approximately 2% of the population, with complete patency being observed in less than 0.1% of the population. The condition is most frequently observed in male infants.[3] Clinical presentation usually includes umbilical discharge, abdominal pain, hernia, rectal bleeding, and umbilical polypoidal or tubular outpouching with occasional presentation as intestinal obstruction due to prolapsed bowel. A fluoroscopic sinogram is the diagnostic modality of choice in OMD anomalies.[4] Differential diagnoses include umbilical granuloma and urachal remnants, which may present with similar symptoms as OMD anomalies.[4]

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We report a rare case of a 10-day male neonate who presented with umbilical swelling and discharge.

CASE REPORT

A 10-day male neonate, born through spontaneous vaginal delivery at the 38th week of gestation with normal anthropometric measurements, presented to the radiology department with an umbilical swelling. He was first born of non-consanguineous parents and was partially vaccinated as per the expanded program of the immunisation schedule. Presenting complaints included yellow-coloured discharge from the umbilicus since birth. There was also documented history of edematous swelling of the umbilical cord along with meconium discharge.

The physical examination revealed a tubular structure protruding through the umbilical orifice with an oozing yellow-coloured discharge through it (Figure 1A). We proceeded with sinogram fluoroscopic study to delineate the communication of umbilical outpouching to internal abdominal organs. Omnipaque contrast was administered into the umbilical herniated structure *via* a butterfly cannula. Pulsed fluoroscopy was used to reduce the radiation dose rate. The contrast was seen outlining the umbilical tubular structure with opacification of small bowel loops suggesting fistulous communication between the umbilicus and ileal loops, *i.e.*, OMF (Figures I B & C). Complimentary CT cuts were taken at the same moment to rule out associated complications *i.e.*, hemorrhage, inflammation or perforation. CT showed a polypoidal, elongated, soft tissue attenuation structure protruding through the umbilical orifice measuring 11.2 × 9.8 × 18.5 mm (Cranio-caudal × Antero-posterior × Transverse dimensions) (Figure I D). Contrast injected through

the aforementioned structure was seen passing into the ileal loops. The rest of the baseline investigations were unremarkable. The infant underwent exploratory laparotomy the next day in which resection of the fistulous tract and end-to-end gut anastomosis were performed. The child remained stable afterwards.

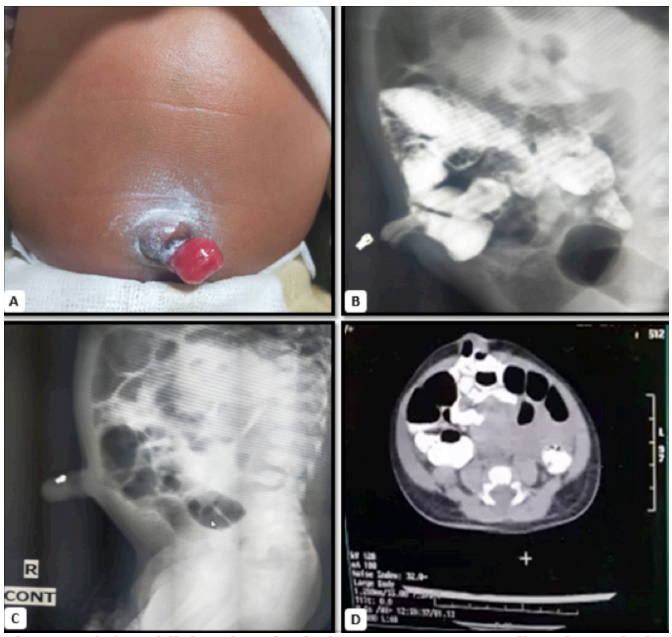

Figure 1: (A) Reddish-colored tubular structure protruding through the umbilical orifice. (B) Administration of contrast delineates the fistulous tract between the umbilical orifice and ileal gut loops. (C) Control spot film showing soft tissue area protrusion at the level of umbilicus. A marker was placed adjacent to this. (D) Computerised tomography axial section at the level of the umbilicus demonstrates the passage of contrast administered through umbilical orifice into ileal gut loops suggestive of fistulous communication between the umbilical orifice and ileal gut loops.

DISCUSSION

The OMF results from a defect in the involution of the vitelline duct occurring between the seventh to tenth week of intrauterine development. Defects in the OMD may result from a partial obliteration of the vitelline duct (Meckel's diverticulum, umbilical sinus, vitelline cyst, or fibrous cord connecting the umbilicus to the intestine) or complete persistence of the vitelline duct resulting in OMF.[4,5]

Kadian *et al*. in their study concluded that OMD anomalies have a male-to-female ratio (M:F) of 4.3:1 with mean age of presentation of 2 months.[5] Rege *et al.* reported a case of an adult patient with patent vitello-intestinal duct and urachus identified intra-operatively on diagnostic laparoscopy done for umbilical hernia repair.[6]

Because of extremely rare occurrence, very little is known about OMF. Therefore, further multi-centre studies are required to have better insights of this phenotype regarding pathogenesis, clinical course, and prognosis. Moreover, the unusual presentation can be a feature that demands a high index of suspicion for OMF in neonates presenting with umbilical swelling or discharge. This case is described to highlight the presence of this extremely rare phenotype to raise a high index of suspicion and awareness among healthcare providers.

PATIENT'S CONSENT:
Written informed consent was obtained from the patient's parents to publish this case report.

COMPETING INTEREST:
The authors declared no competing interest.

AUTHORS' CONTRIBUTION:
AT: Designed the study, analysed it, and drafted the manuscript.
MII: Revised it critically and supervised the study.
TN: Revised it critically and proofread.
MZA: Contributed to data collection and drafted the manuscript.
HA: Contributed to data collection and proofread.
All the authors have approved the final version of the manuscript to be published.

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