



Case report

Perforated Meckel's diverticulum in omphalocele in a newborn: A case report of an uncommon presentation from northern Tanzania

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ABSTRACT

Introduction and importance: The concurrent existence of Omphalocele and Meckel's diverticulum is not unheard of but is relatively uncommon. A few cases of their coexistence have been reported. Due to the uncommon dual presentation, it is easy to delay or even miss the diagnosis, delaying management. Meckel's diverticulum should be considered if there is a bowel opening on an omphalocele.

Case presentation: Herein we present a newborn male baby who was referred to us presenting with an omphalocele that was leaking faeces. The baby also had a cleft lip and palate. He was born at term to a 30-year-old mother whose pregnancy was otherwise normal. The fistulated omphalocele was surgically repaired, and the child continued to do well.

Clinical discussion: Omphalocele and Meckel's diverticulum are both relatively rare congenital malformations that are uncommonly present together. Other congenital malformations can be associated; hence thorough investigations should be carried out when resources are available. The search for associated malformation should not delay the management of the pathology as it can have serious consequences on the health and outcome of the child.

Conclusion: Fistulation of Meckel's diverticulum on an Omphalocele is rare. Treatment involves surgical resection and repair. Though other co-morbidities should be investigated, investigation for cause and other co-morbidities should not delay surgery.

1. Background

Omphalocele is a rare anterior abdominal wall malformation with an incidence rate of 1 in 4000 live births and was first described in 1634 [1]. Meckel's diverticulum is an uncommon congenital defect of the gastrointestinal tract, and reports of the concurrent pathologies are very few [1]. Herein we present a case of perforated Meckel's diverticulum in an Omphalocele in a newborn who was successfully managed collaboratively at our tertiary health care centre in northern Tanzania despite being in a resource-limited setting. Medical personnel should have a high index of suspicion of Meckel's diverticulum when Omphalocele is seen in neonates.

This work has been reported in line with the SCARE 2020 criteria [2].

2. Case presentation

The baby presented to our hospital three days after delivery with umbilical swelling, cleft lip and palate, and passing stools per umbilicus since birth. The informant was the mother who reported term delivery after an uneventful pregnancy. She denied any history of fevers.

The baby was delivered at a local health centre by spontaneous vaginal delivery, weighing 2.1 kg and cried immediately after birth. Antenatally, the mother booked at four months of gestation age and visited antenatal clinic five times. She received all supplements as per local guidelines. She tested negative for HIV, and VDRL was non-reactive. She was normotensive throughout her antenatal visits.

At age 30, this was the mother's 2nd delivery. Her firstborn was seven years old, alive and well. She denied alcohol and cigarette use.

Abbreviations: ANC, antenatal clinic; ECHO, echocardiogram; HIV, human immunodeficiency virus; NGT, naso-gastric tube; VDRL, venereal disease research laboratory.

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An initial examination found the baby awake, calm, febrile (38.2 °C), with an NGT in situ (placed at referring facility). The baby had a respiratory rate of 64 breaths per minute and was saturating at 98% on room air. His heart rate was 144 beats per minute, and his random blood glucose was 3.6 mmol/L. The anterior fontanelle measured 5 by 2.5 cm. On the left side of the face, the baby had a cleft lip and palate (Fig. 1).

On the abdomen, the baby had an omphalocele measuring 5 by 5 cm with a perforation on it that was leaking stool. The omphalocele had been covered with moist sterile gauze. The abdomen felt soft, and no enlarged organs could be palpated. The baby had normal male external genitalia with both testicles palpable in the scrotum (Fig. 2). Our primary diagnosis was fistulated Omphalocele in a baby with Patau syndrome. Our other diagnoses were neonatal sepsis, cleft lip and cleft palate.

The baby was nursed in the neonatal unit, fed by NGT to prevent aspiration and was kept on intravenous antibiotics. Complete blood count revealed a haemoglobin level of 19.1 g/dl, normal white cell count of $14.94 \times 10^9/L$, and platelet count of $161 \times 10^9/L$. His serum potassium was 5.99 mmol/L, sodium 141.04 mmol/L, creatinine 135 $\mu\text{mol/L}$ and urea 9.91 mmol/L. Echocardiogram was normal. The child was started on a regiment of ceftriaxone and metronidazole and received a stat dose of vitamin K.

The patient's workup was started on the day of admission. Within 24 h of admission (day 4 of life), the child was taken for a laparotomy by the consultant pediatric surgeon. The abdomen was opened through a transverse incision about the omphalocele. The small bowel, approximately 20 cm proximal from the ileocecal junction, was found trapped in the omphalocele and fistulated (Meckel's diverticulum). The bowels were released, and an 8-centimetre segment was resected. End-to-end ileo-ileal anastomosis was done (Fig. 3). Patency was established. The abdomen was closed in layers, and the omphalocele was repaired. The baby was then sent back to the neonatal ward to complete the course on antibiotics. Feeding was initiated 12 h after the surgery starting at 5mls of expressed breast milk supplemented by intravenous 10% dextrose. Oral feeds were gradually increased with subsequent feeds as the child demonstrated tolerance.

The baby continued to do well and was discharged six days after surgery, having tolerated oral feeds and passing stools. Review at surgical outpatient unit 6 weeks post-discharge found him to be well with good weight gain. Histological examination of the resected segment of



Fig. 2. Photograph showing fistulated omphalocele (arrow).

the bowel showed normal intestinal tissues. Surgery to repair the cleft lip and palate was scheduled after three months.

3. Discussion

In fetal life, between the 5th and 7th weeks of gestation, the midgut and hindgut herniate into the umbilicus as they grow because the abdominal cavity is relatively small. By the 11th week, the guts return into the abdominal cavity. Failure to do so produces an omphalocele or gastroschisis [1]. Environmental factors and young maternal age appear to contribute to gastroschisis, while advanced maternal age and genetic factors contribute to omphalocele. Gastroschisis has fewer associated anomalies (8%) compared to omphalocele (35%) [1]. It was evident in our case that the child had other phenotypic abnormalities. Unfortunately, we could not do genetic analysis to confirm or rule out genetic anomalies associated with Omphalocele, e.g. Trisomy-21, Marshall-Smith syndrome and Meckel-Gruber syndrome [3].

Omphalocele is an uncommon midline defect occurring in 1 in 4000 to 6000 live births [4]. Small omphalocele size is associated with fewer cardiac anomalies but a higher prevalence of gastrointestinal anomalies such as ileal or colonic atresia and vitello-intestinal duct abnormalities, as evident in our case [4]. Other anomalies include cleft lip or palate, genital or diaphragmatic malformations, central nervous system malformations, and genitourinary malformations [4,5].

Meckel's diverticulum is a rare condition that occurs at an annual frequency between 0.6% and 4% [6]. It is a result of incomplete regression of the omphalomesenteric duct and cannot be diagnosed prenatally unless it is dilated [1]. As shown in our case, it can be attached to the sac of the omphalocele that can be another factor that prevents bowels from returning to the abdominal cavity intrauterine [1]. Charki et al. state in their report that Meckel's diverticulum was found in 16% of the cases of omphalocele with a higher incidence in smaller (28%) compared to larger omphalocele (4%) [4]. We thought that the diverticulum fistulated because of the increased intraluminal pressure of the small bowel as the neck of the diverticulum was narrow, leading to the build-up of contents hence fistulating.

The management involves resection of the loop of the intestines, including the Meckel's diverticulum, with end-to-end anastomosis to maintain gastrointestinal continuity [4,7]. The repair of the abdominal wall can be done in a staged manner, as reported by Jin et al. [1]. Due to the small size of the omphalocele in our case, repair of the anterior abdominal wall was done in the same sitting to avoid second anaesthetic



Fig. 1. Photograph showing left-sided cleft lip.

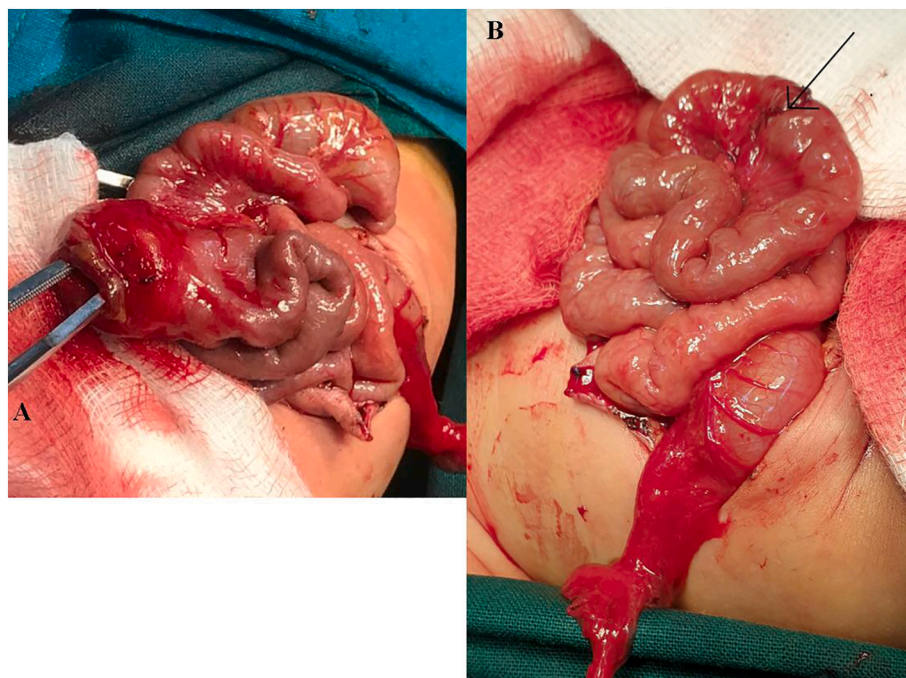


Fig. 3. A: Fistulated Meckel's diverticulum. B: End-to-end anastomosis (arrow) post-resection.

and surgery exposure. Larger omphaloceles are challenging for primary closure due to high morbidity and mortality, such as abdominal compartment syndrome and respiratory compromise; however, the introduction of silo or absorbable synthetic mesh repair has provided a positive outcome [8,9]. In low-resource settings like ours, conservative management of large omphaloceles can be sought using topical alcohol or vacuum-assisted closure method that promotes escharification and epithelization of the omphalocele to form a ventral hernia. However, if the sac ruptures, then surgery is an absolute indication [8–10].

The fact that the case could be managed both surgically and supportively to end in a positive outcome speaks to the team's strengths that managed this child. However, weaknesses such as the pathology not being identified beyond it being a congenital malformation in the centre where the child was delivered. This speaks towards the low awareness of such pathologies and hence the need for case reports such as this to educate the medical community. Genetic testing would have also been ideal for identifying the possible syndrome that resulted in the combination of pathologies that the baby displayed, but this was beyond the reach of the centre and the patient.

4. Conclusion

Fistulisation of Meckel's diverticulum in an omphalocele is rare. Surgical resection of the diverticulum and repair of the anterior abdominal wall remains the standard of treatment.

Consent

Written informed consent was obtained from the child's mother for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Ethical approval was obtained from the department of General surgery and department of Pediatrics and Child health, KCMC Hospital.

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Author contributions

Jay Lodhia – conceptualization and writing of the script
 Mathayo Shadrack – reviewed medical records and writing of the script
 David Msuya – lead surgeon and reviewed medical records
 Levina Msuya – reviewed medical records
 Rune Philemon – reviewed the records and conceptualizing of script

Guarantor

Jay Lodhia – Corresponding author.

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N/A.

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Declaration of competing interest

The authors declare they have no competing interests.

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