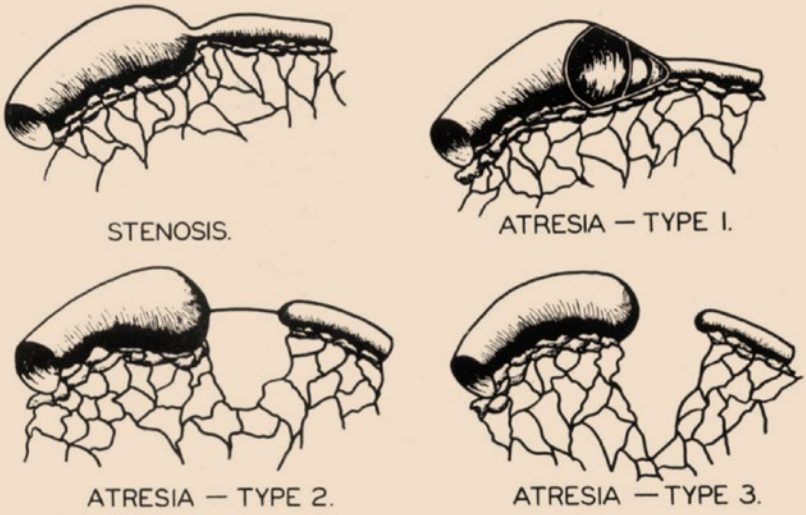
SWISS SOCIETY OF NEONATOLOGY

A surprising result of a prenatally suspected ovarian cyst

August 2019



Preisser P, de Bernardis G, Scheibner K, Grobholz R, Meyer P, Department of Neonatology (PP, MP), Department of Pediatric Surgery (DBG), Children's Hospital of Aarau, Department of Obstetrics and Gynecology (SK), Department of Pathology (GR), Cantonal Hospital of Aarau, Aarau, Switzerland.

Title figure:
Classification of intestinal atresia
(Source: Louw JH. Moynihan Lecture, April 24, 1959).

CASE REPORT

This was the third pregnancy of a 39-year-old woman. During routine follow-up at 36 weeks of gestation, an intraabdominal mass measuring 5 × 4 cm was noted on fetal ultrasound examination. The finding was confirmed one week later by an experienced examiner; appearance and size of the mass had not changed (Fig. 1).

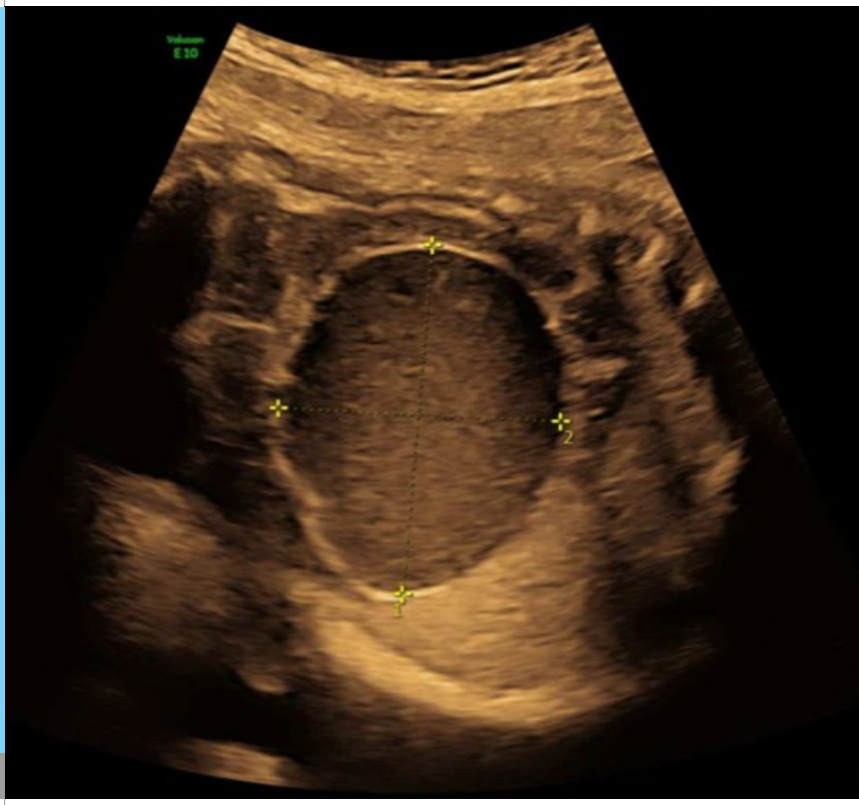


Fig. 1

Prenatal ultrasound examination at 36 weeks of gestation: thin-walled intraperitoneal cyst (5.4 × 4.3 cm) in the left lower hemiabdomen with suspected intracystic hemorrhage.

The ultrasound scan revealed a thin-walled, intraperitoneal cyst with signs of an intracystic hemorrhage in the left lower hemiabdomen. There was no evidence of connections to the bladder or intestine; however, mild polyhydramnios was documented. An ovarian cyst was felt to be the most likely diagnosis.

The baby's postnatal management was discussed by an interdisciplinary team, consisting of neonatologists, pediatric surgeons and feto-maternal specialists. The baby was to be examined clinically by a neonatologist, and an abdominal ultrasound should be performed within the first days of life. Postnatal admission to the neonatal unit was not deemed necessary if adaptation and clinical exam were normal.

At 39 1/7 weeks, the female infant was born via normal vaginal delivery and adapted well with Apgar scores of 8, 10, and 10 at 1, 5, and 10 minutes, respectively. The arterial umbilical cord pH value was 7.41. Birth weight was 3280 g (P40).

The baby's clinical examination after birth was unremarkable without a palpable abdominal mass, and the initial postnatal course was uneventful. She was breastfeeding well and passed meconium and urine within the first 24 hours after birth.

On the third day of life, before the prenatally planned ultrasound examination was performed, she presented

with biliary vomiting and was admitted to the neonatal intensive care unit. On admission, she was clinically well, and her vital signs were within normal limits. The abdomen was distended, and mass was now felt in the left abdomen; in addition, abnormal bowel sounds were noticed. A babygram revealed dilated intestinal loops and absence of gas in the rectum (Fig. 2).

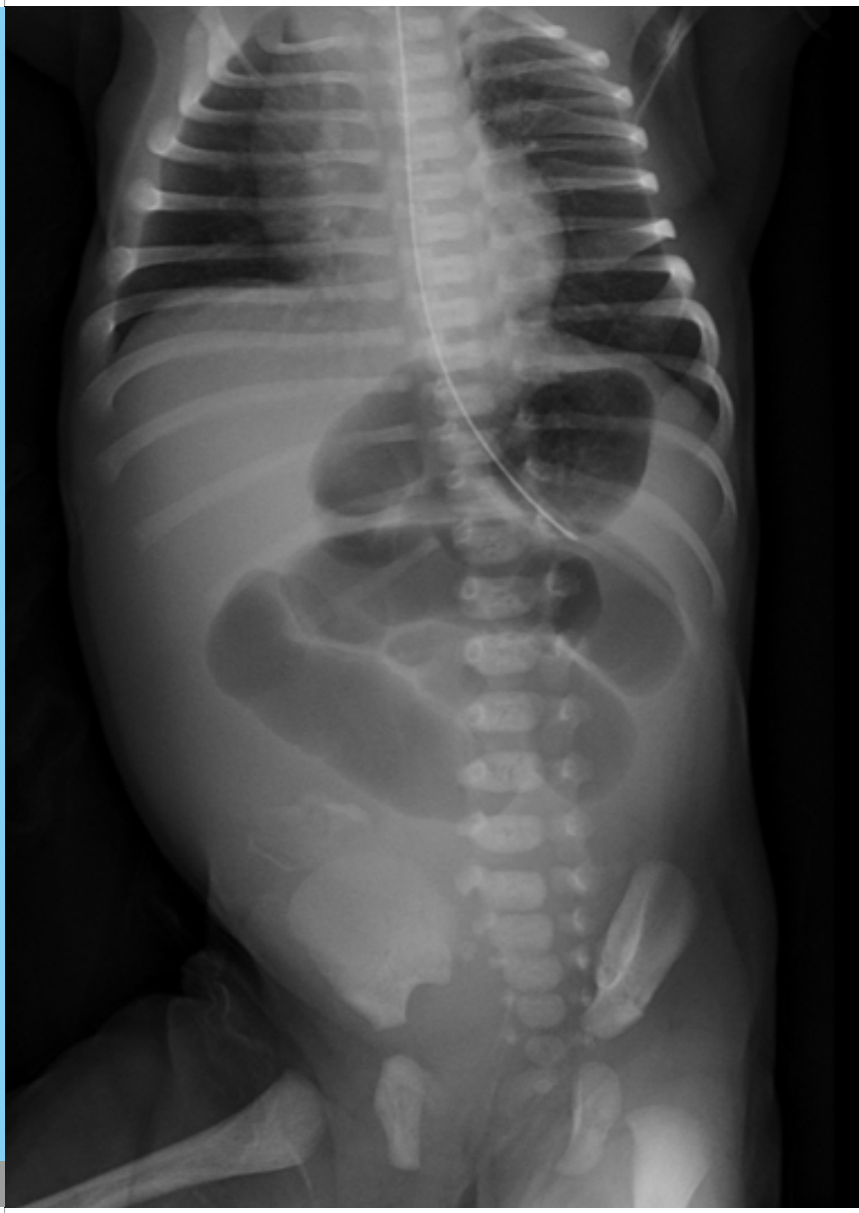


Fig. 2

Babygram obtained after the onset of bilious vomiting on DOL 3: abnormal gas pattern with massively distended bowel loops and absent air in the rectum.

Ultrasound examination of the abdomen was difficult due to massively dilatated loops of bowel in the upper abdomen. Nevertheless, a cyst could be demonstrated in the right lower abdominal quadrant measuring 3.7 × 5.0 × 4.3 cm. The patient's history and the X-ray findings were consistent with a mechanical ileus. Because of the sonographic appearance of the cyst with typical egg shell calcifications, meconium peritonitis was suspected. Therefore, an exploratory laparotomy was felt to be indicated.

Intraoperatively, a cyst with a diameter of 4 cm, tightly attached to a small bowel segment was found. At the site where the cyst was attached, the continuity of the small bowel was interrupted by a fibrotic string over a distance of about 2 cm, consistent with a type II bowel atresia (Grosfeld's classification). The cyst and the atretic bowel segment were resected (Fig. 3), and bowel continuity was restored by an end-to-end anastomosis despite the fact that there was noticeable atrophy of the bowel distal to the atretic segment. Other bowel obstructions or malformations, such as Ladd's bands or malrotation were excluded.

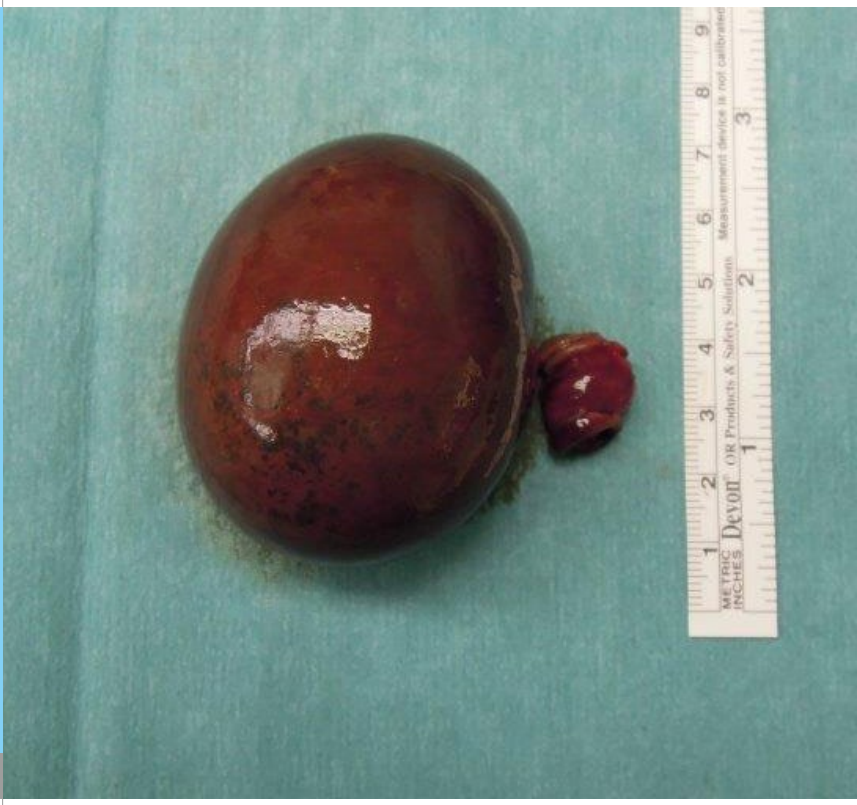


Fig. 3

Resected cyst and atretic bowel segment (ileum).

The histopathologic examination of the resected cyst failed to identify any bowel tissue within the cyst but demonstrated remnants of paramesonephric ducts in the cyst wall. Given the fact that the cyst was connected to the atretic part of the bowel (a finding that has never been described for paramesonephric duct remnants in the literature), it was believed that this was a casual finding rather than an etiologic explanation.

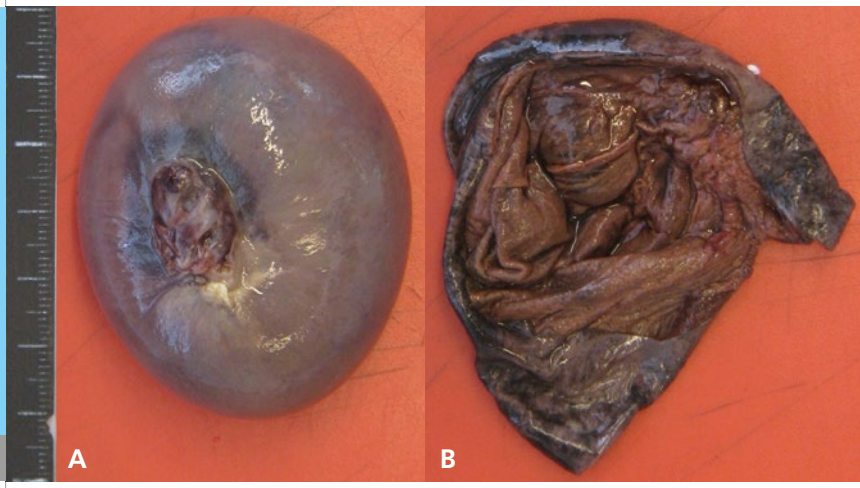


Fig. 4

Macroscopic appearance of the cyst (A: prior to incision; B: after incision).

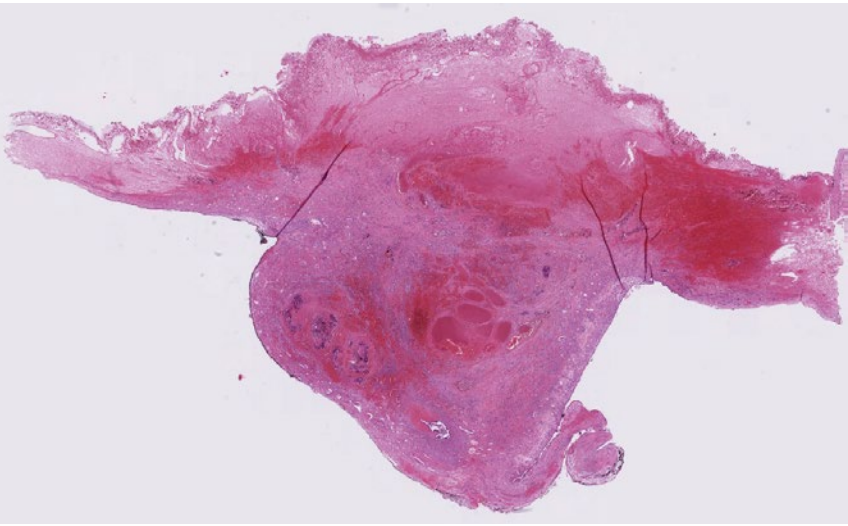


Fig. 5

Histology: overview cyst wall with hemorrhage (magnification x10).

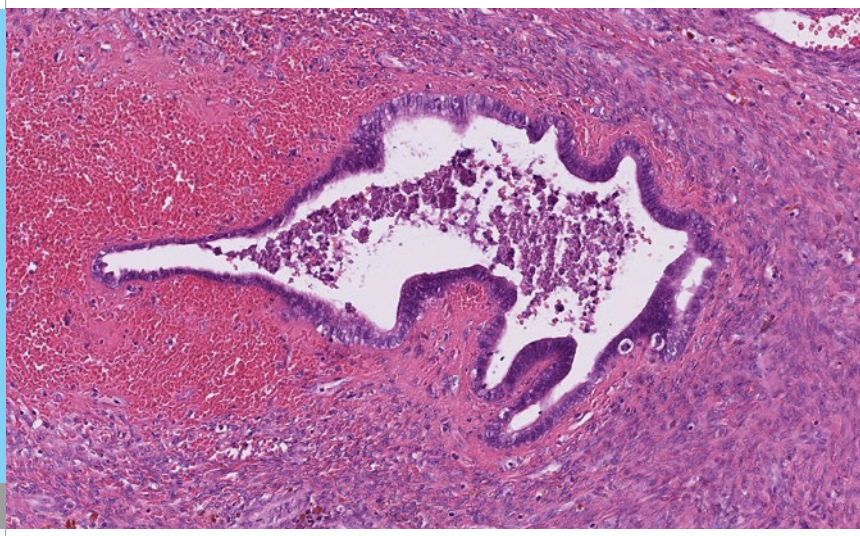


Fig. 6

Histology: mesonephric remnants in cyst wall (magnification x100).

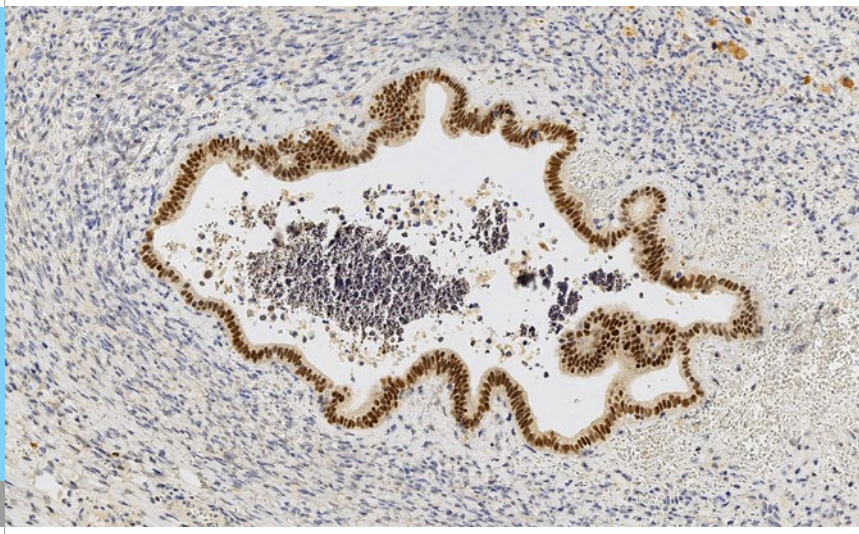


Fig. 7

Immunehistochemistry: expression of transcription factor PAX2 in epithelium of mesonephric remnants.

Enteral feedings were introduced on the first post-operative day and gradually increased. They were well tolerated, and, 16 days after surgery, the infant was on full enteral feeds, had displayed good weight gain and could be discharged home.

DISCUSSION

There is a wide spectrum of differential diagnoses for congenital abdominal masses, including renal, genital and gastrointestinal anomalies. Among these, anomalies of the urinary tract belong to the most common causes of cystic abdominal masses, followed by dilated bowel, ovarian cysts, enteritic duplication cysts, hepatic und biliary cysts, meconium pseudocysts and mesenteric cysts (1). Marchitelli et al. showed an accuracy of 90 % for the diagnosis of fetal cystic lesions made by ultrasound (2).

Ovarian cysts are detected prenatally in about 1/1000 fetuses (3). A false-positive rate of 7.5 % has been described by Bascietto et al (4). Both pre- and postnatal management of ovarian cysts depends on the size, growth rate and potential complications. It includes antenatal aspiration, postnatal surgery with resection or a wait-and-see approach with sonographic follow-up due to the possibility of spontaneous resolution (5 – 7).

While ovarian cysts often regress spontaneously, other lesions generally require surgery. Therefore, determination of the origin of the cyst is important to define a schedule for prenatal follow-up and to assess the likelihood of a postnatal surgical intervention. Obviously, it is also relevant for prenatal counselling.

In our case, the postnatal clinical presentation with biliary vomiting indicated an ileus related to the

suspected ovarian cyst. In general, smaller cysts remain free of symptoms, but in case of a large size of the cyst, a torsion or a rupture, symptoms of an ileus can occur. Irrespective of the suspected diagnosis, a surgical intervention must be considered and may reveal, as demonstrated by our case, a completely different etiology.

REFERENCES

1. McEwing R, Hayward C, Furness M. Foetal cystic abdominal masses. Australas Radiol 2003;47:101 – 110 *(Abstract)*

2. Marchitelli G, Stirnemann J, Acanfora MM, Rousseau V, Salomon LJ, Ville Y. Prenatal diagnosis of intra-abdominal cystic lesions by fetal ultrasonography: diagnostic agreement between prenatal and postnatal diagnosis. Prenat Diagn 2015;35:848 – 852 *(Abstract)*

3. Tyraskis A, Bakalis S, David AL, Eaton S, De Coppi P. A systemic review and meta-analysis on fetal ovarian cysts: impact of size, appearance and prenatal aspiration. Prenat Diagn 2017;37:951 – 958 *(Abstract)*

4. Bascietto F, Liberati M, Marrone L, et al. Outcome of fetal ovarian cysts diagnosed on prenatal ultrasound examination: systemic review and meta-analysis. Ultrasound Obstet & Gynecol 2017;50:20 – 31 *(Abstract)*

5. Trinh TW, Kennedy AM. Fetal ovarian cysts: review of imaging spectrum, differential diagnosis, managment, and outcome. Radiographics 2015;35:621 – 635 *(Abstract)*

6. Turgal M, Ozyuncu O, Yazicioglu A. Outcome of sonographically suspected fetal ovarian cysts. J Matern Fetal Neonatal Med 2013;26:1728 – 1732 *(Abstract)*

7. Tyraskis A, Bakalis S, Scala C, et al. A retrospective multicenter study of the natrual history of fetal ovarian cysts. J Pediatr Surg 2018;53:2019 – 2022 *(Abstract)*

concept & design by mesch.ch

SUPPORTED BY

VIFOR PHARMA

CONTACT

Swiss Society of Neonatology
www.neonet.ch
webmaster@neonet.ch