Reminder of important clinical lesson

Management of ovarian cysts during infancy: autoamputation presenting as a possible pitfall

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SUMMARY

The widespread use of routine antenatal ultrasound has relatively increased the frequency of intrauterine diagnosis of ovarian cysts. In utero adnexal torsion may present with subsequent autoamputation in some of these lesions. Prenatal and postnatal ultrasonographic findings, however, may not always be relevant in making the correct diagnosis. The authors report on two cases with prenatally-diagnosed hypoechogenic cystic masses. The cysts failed to resolve after a period of conservative management. A laparoscopic approach revealed ovarian autoamputation presenting as cystic mass. Preoperative ultrasound, abdominal CT and MRI failed to detect the presence of autoamputation. The present report discusses the possibility of an otherwise silent ovarian autoamputation, which may necessitate laparoscopic intervention for correct diagnosis, in neonates presenting with persistent ovarian cysts.

BACKGROUND

The treatment of ovarian cysts is not standardised, but based on the size and appearance of the cyst on sonographic examination. Some authors report that complex but asymptomatic neonatal ovarian cysts have a natural tendency to involute spontaneously and, therefore, recommend conservative management with clinical and sonographic monitoring.1-6 The ability of antenatal ultrasound to determine long term outcome of torsion or spontaneous haemorrhage into follicle cysts is challenged, and prenatal and postnatal ultrasound findings may not always be relevant in making the correct diagnosis. A sonographically persistent ovarian cyst may be free floating in the peritoneal cavity with no connection to the pelvis, or may parasite omental circulation and simulate an omental cyst.5,7-9

We aim to highlight the possible presence of an otherwise silent ovarian autoamputation in neonates presenting with persistent ovarian cysts. We believe that laparoscopic intervention is necessitated for exact diagnosis, under suspicion of autoamputation.
CASE PRESENTATION

Case 1

A 7-month-old girl was referred to our clinic with a 35x28x29 mm left adnexal cystic mass, which failed to resolve after a period of conservative management. It was first detected at 34 weeks of gestational age, measuring 32x33x40 mm with an echogenicity mimicking solid appearance (fig 1). Post delivery ultrasound (US) at 37 weeks of gestational age reported a left adnexial cystic mass measuring 43x36x41 mm, with smooth surface and septations inside. Physical examination revealed a smooth, mobile, lower abdominal non-tender mass on admission. On admission, haematological and biochemical investigations were within normal limits with normal β-human chorionic gonadotropin (hCG) and α-fetoprotein (αFP) levels. Abdominal CT reported a 5 cm cystic mass with solid components in left adnexial area.

Figure 1 Prenatal ultrasound showing an abdominal mass measuring 32x33x40 mm at 34 weeks of gestational age. DLK, spleen; MASS, mass; MIDE, stomach; SOL BOB, left kidney.

Case 2

A term newborn was referred to our clinic with bilateral adnexal cystic masses (left 48x43 mm, right 38x30 mm) detected during prenatal US. An MRI study revealed bilateral haemorrhagic cysts (left 30x50x70 mm, right 30x25x40 mm) when she was 12 days old. Sonographic findings showed persistence of adnexal cystic masses (left 39x55x50 mm, right 30x37x31 mm). The right ovarian cyst continued to persist while the left cyst decreased to 15x12x12 mm in diameters. Serum αFP levels decreased to normal from over 30,000, and β-hCG was within normal limits during follow-up. CT obtained at 9 months of age showed a cystic lesion 30x40x35 mm in diameter with pelvic median line localisation and a 15 mm calcification area on the anterior wall. Physical examination showed a palpable, mobile, semisolid mass 30–40 mm in size.

DIFFERENTIAL DIAGNOSIS

US and CT may fail to detect the exact diagnosis in patients with ovarian cysts.
TREATMENT
We successfully treated both children via a laparoscopic approach.

Case 1: the laparoscopic approach revealed a freely floating soft, rounded and brown cyst with a blind ending left fallopian tube (fig 2). The right ovary appeared normal and was left in situ. The cyst was successfully removed through the 5 mm umbilical port following drainage of its haemorrhagic content.

![Figure 2](image.png)

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Figure 2 Laparoscopic view of the blind ending left fallopian tube, grasper is pointing toward the uterus, the right ovary can be seen under the grasper.

Case 2: the laparoscopic approach revealed ovarian autoamputation with a free floating cyst with vessel resembling attachments originating from the greater omentum, a blind ending right fallopian tube and a normal left ovary. The cyst was removed successfully as described in the first case.

OUTCOME AND FOLLOW-UP
Case 1 was discharged home on the second postoperative day. Histopathology revealed a cyst with haemorrhage and areas of infarction.

Case 2 was discharged on third postoperative day. Histopathology revealed a cyst with haemorrhage, areas of infarction and calcification.

Both patients are doing well.

DISCUSSION
The incidence of neonatal ovarian cysts is reported as 34%. On the basis of their sonographic features, these cysts are divided into two types: simple and complex, also known as complicated. Simple ovarian cysts tend to resolve spontaneously, which is known to occur in 25% to 50% of cases in the first year of life. Some, however, may fail to regress and become complicated.
Prenatal ultrasonography is a reliable method in determination of the abdominal masses, especially cystic lesions such as ovarian, kidney, mesenteric, urachal and intestinal duplication cysts. MRI is a more suitable radiological examination for achieving the final diagnosis and characterising the content of the lesion when compared to CT. Complications, however, may be misdiagnosed.

The most common reported complication is torsion, bearing an incidence of 50% to 78% of all neonatal ovarian cysts. Spontaneous ovarian torsion may occur prenatally or in neonatal period. The risk of torsion is said to correlate with the size of the cyst. The risk is higher for cysts greater than 4 to 5 cm in size.

Surgical removal of the mass is most often recommended in simple cysts more than 5 cm in diameter and complex cysts showing debris/fluid level, septa, a retracting clot or mural nodule, or echogenicity mimicking a solid appearance. Persistence, presence of calcified components within the cyst wall and evidence of intracystic bleeding prompted intervention in our cases. Published reports disclose that some type of oophorectomy or salpingo-oophorectomy is performed in most of these patients, besides the strong recommendation toward preservation of the ovarian tissue.

Autoamputation is a very rare complication. This process may occur asymptotically and may lead to congenital absence of the ovary or a free floating, often partially calcified cystic mass in the abdomen, as in our first case. US and CT failed to detect torsion and autoamputation in our patients, as in the reported literature. The case report presented by Peh et al is another example of late presentation in a young woman with a missed diagnosis. Our second case presented a vessel-like attachment originating from the greater omentum (fig 3). Curarrino has reported a similar case where the cyst seemed to parasite omental circulation and simulated an omental cyst at surgery. Our patient may support the suggestion that the gonad was still in its initial position and had not yet descended to the pelvis during the pathological process.

Figure 3 Autoamputated right ovarian cyst, with vessel resembling appearance originating from the greater omentum, showing additional attachments to the anterior abdominal wall.
Laparoscopic management provides correct diagnosis in newborn ovarian pathologies and allows therapeutic interventions including aspiration, fenestration, cyst excision, or oophorectomy. The magnitude of the cyst is not a contraindication, since laparoscopic cyst puncture and aspiration allows sufficient reduction in size. The possibility of an otherwise silent ovarian autoamputation may necessitate laparoscopic intervention for correct diagnosis in neonates presenting with persistent ovarian cysts.

**LEARNING POINTS**

- Differential diagnosis is important among simple and autoamputated ovarian cysts.
- Ultrasound and CT may fail to detect the exact diagnosis in patients with ovarian cysts.
- A laparoscopic approach in infants with similar diagnosis is feasible and should be regarded as the first line of treatment.
- Our second case may be an example of what might happen if early ovarian torsion is present before gonadal descent takes place, or may be a clue of an abnormal embryonic development of the primitive gonad.

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**Patient consent:** Patient/guardian consent was obtained for publication.

**REFERENCES**


