Paediatric cystic ovarian torsion masquerading appendicitis

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SUMMARY
A 7-year-old girl presented with a 2-day history of right iliac fossa pain, fever and elevated inflammatory markers. Clinical examination supported a diagnosis of appendicitis. The patient was taken to theatre for an open appendicectomy the following morning. Intraoperatively, a right-sided ovarian haemorrhagic cyst with 360 degrees torsion was discovered. The ovary was torted along with the cyst. Both were detorted and the abdomen was closed. The patient was discharged 48 hours later, with gynaecology outpatient follow-up 6–8 weeks later. Paediatric ovarian torsions caused by a haemorrhagic cyst greater than 2 cm are rare. Here, we discuss an atypical presentation of ovarian torsion and how the clinical presentation can mimic appendicitis.

BACKGROUND
In the paediatric population, torsion of the ovary is rare with an incidence of around 5 in 100 000.1 While ovarian cysts measuring 1 cm or less have a prevalence in the prepubertal population of 2%–5%, cysts greater than 2 cm are considered to be rare.2 Some cysts are hormonally active and facilitate the development of sexual characteristics.3 Cysts greater than 2 cm require follow-up until regression is seen due to their risk of torsion.

Ovarian torsions usually present with a sudden onset of abdominal pain and may be a differential diagnosis of an acute surgical abdomen. Torsion usually results from twisting of the ligamentous support that results in venous congestion.1–3 The condition has previously been managed with an oophorectomy; however, a systematic review has shown detorsion alone to be sufficient, with promising results for future fertility.1

CASE PRESENTATION
A 7-year-old girl presented to the accident and emergency department in the evening with a 2-day history of worsening right-sided abdominal pain. The pain was non-migratory in nature and was associated with anorexia, a single episode of diarrhoea and vomiting. This was the first time she had experienced this pain. The patient was fit and well with no other medical conditions. She was premenarcheal. Her mother gave an uneventful perinatal history and family history.

On assessment by the surgical team, the patient was found to be feverish with a temperature of 38.1°C. She displayed generalised tenderness over her lower abdomen, but particularly in the right iliac fossa. Her bloods revealed an inflammatory response with a white blood cell (WBC) count of 19 x 10⁹/L and C reactive protein of 62 mg/L. The patient was started on intravenous antibiotics. As the patient remained clinically stable overnight, a plan was made to perform an open appendicectomy the following morning.

Intraoperatively, she was found to have an appendix adhering to the posterior aspect of the caecum, which looked macroscopically normal. While no free fluid was identified, mesenteric lymph nodes of the small bowel were identified with no Meckel’s diverticulum. A decision was made to remove the appendix.

Before closure, a limited inspection of the pelvis was performed through the right iliac fossa wound. This revealed a suspicious black lesion and so the wound was extended to allow further exploration. The patient was found to have a large haemorrhagic torted ovarian cyst measuring 7x3 cm, which ruptured on mobilisation. The attached ovary appeared ischaemic (figure 1). The cyst had torted through a full 360º. The ovary’s appearance did not change following detorsion. After detorting the cyst, senior gynaecology input was sought. The advice was that, in spite of the appearance of the ovary, there was potential for it to recover and retain function, and so both the ovary and the cyst were carefully placed back into the pelvis. The wound was closed as per normal.

Over the following 24 hours, the WBC normalised and the patient improved clinically. She was discharged home and has recovered well.

INVESTIGATIONS
The patient had a urinary microscopy culture and sensitivity, which was unremarkable.

No imaging was conducted.

OUTCOME AND FOLLOW-UP
The patient has recovered well following her surgery. The patient had a follow-up ultrasound scan at 3 months, which demonstrated the left ovary measuring 38x17x26 mm and a right ovary measuring 18x10x18 mm with viable ovarian tissue.

DISCUSSION
The case described above is uncommon as both the ovary and associated haemorrhagic cyst of 7x3 cm had torted. Differentiating appendicitis from ovarian torsion is difficult as they present in very similar ways. It is often thought that ovarian torsion is secondary to a cyst or ovarian tumour.1 However, other theories have been proposed to
explain torsion in a regular ovary, including excessive movements secondary to a predisposing long utero-ovarian ligament.3

Appendicitis is considered a clinical diagnosis, with blood tests and imaging playing a supportive role. In our case, there was enough clinical suspicion to warrant early surgical intervention. While an ultrasound scan may have diagnosed the true pathology preoperatively, there was a low clinical suspicion of a gynaecological pathology in a prepubertal patient. Research suggests sonography alone to be inconclusive in this age group.6 Doppler ultrasound scans should be considered as they offer the greatest sensitivity and specificity compared with MRIs and CT.6,7

Traditional surgical intuition would be to remove a structure that appeared ischaemic and non-viable. However, current evidence suggests that the cyst and ovary should be left in situ as there is potential for the ovary to function as normal.1 The necrotic appearance of the ovary is more a consequence of venous congestion as opposed to arterial insufficiency. Therefore, detorsion alone may provide a good chance of recovery. Long-term follow-up of adult females following detorsion of an ovary has shown folliculogenesis on ultrasonography and successful pregnancies.3 Traditionally, oophorectomies have been performed due to the potential of missing a malignant cause. However, further evidence is required to support this.1,8

Resected ovarian specimens have shown the presence of viable parenchyma despite their necrotic appearance advocating for the use of ovarian sparing surgery. Laparoscopic surgery can be used for ovarian cysts measuring less than 7.5 cm and a laparotomy for those that are larger.5 The initial approach should be for detorsion alone. The patient must be followed up with regular ultrasound scans. Absence of cyst regression warrants further investigation with CT and tumour markers.9

Regardless of the underlying pathology, it was clear operative management was needed in this case. In the absence of a dedicated paediatric surgical service, an open approach is commonly adopted; especially in smaller patients. This patient may have benefited from a laparoscopic approach, as it would have facilitated quick identification of the underlying pathology and management could have proceeded without extending the incision. Nonetheless, the patient has recovered well and her follow-up imaging have shown promising results for future fertility.

Patient’s perspective

When I was informed that my daughter’s pain had not been caused by appendicitis but due to her right cystic ovary having twisted, although I was relieved the cause had been discovered and rectified, I also felt confused as to how a large cyst had developed in her ovary at such a young age and concerned about her future fertility prospects.

Learning points

► Ovarian torsion should always be considered as a differential diagnosis in all female patients with acute right iliac fossa pain and raised inflammatory markers.
► Ultrasound scans can be particularly useful in the female population. However, it can be overlooked in children with such typical features of appendicitis.
► A laparoscopic approach can help identify pelvic pathology mimicking appendicitis. However, outside of centres for paediatric surgery, often an open approach is preferred, particularly in younger and smaller children.
► Attempts should be made to salvage twisted ovaries regardless of the appearance.
► A follow-up ultrasound must be arranged to ensure regression of the cyst.

Case report

Figure 1 Intraoperative images of torted haemorrhagic cyst and ovary.

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Contributors SK has written the case report with contribution from both SS and DB.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Parental/guardian consent obtained.

Provenance and peer review Not commissioned; externally peer-reviewed.

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