CASE REPORT





Gangrenous Hydrosalpinx with Isolated Fallopian Tube Torsion in Premenarchal Child

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Abstract

Hydrosalpinx induced torsion of the adnexa in a premenarchal child without sexual experience is a rare situation and immediate intervention is necessary. This report describes a 10-year old premenarchal female without a history of sexual intercourse who presented with left lower abdominal pain. Ultrasound imaging revealed a left adnexa with thick-walled cystic lesion measuring $5.1 \times 3.7 \times 2.7$ cm suggestive of hydrosalpinx or para ovarian cyst. Diagnostic laparoscopic surgery showed ischaemic tubular shaped cystic left hydrosalpinx that was twisted along its own axis at the level of isthmus. Laparoscopic left salpingectomy was performed within 24 h of presentation leaving behind a healthy left ovary. Early diagnosis is important in preventing further complications including severe infection and peritonitis and to prevent complications that might affect fertility.

Keywords Hydrosalpinx · Isolated fallopian tube torsion · Adnexal torsion · Premenarchal child

Introduction

Isolated fallopian tube torsion (IFTT) results from the rotation of the fallopian tube about its own axis and leaves the ovary unaffected, without torsion. The incidence of this phenomenon is approximately 1/1,500,000 women [1]. Due to lack of specific clinical symptoms, a clear pre-operative diagnosis is difficult, especially in a case without significant pre-disposing factors. We report the case of a 10-year old premenarchal virgin child with hydrosalpinx induced isolated tubal torsion complicated with gangrene.

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Case Report

A 10-year old child visited the OBGYN OPD and presented with complaints of severe pain over the left lower abdomen radiating to the left upper thigh. The patient's parents requested to visit the OBGYN department due to a similar history in the patient's cousin associated with onset of menarche. She described the pain as dull and colicky with no relieving and exacerbating factors and had no similar complaints before this presentation. She had multiple episodes of vomiting and nausea. There were no symptoms of fever, constipation, diarrhoea, vaginal bleeding or dysuria. Patient had not attained menarche but gives a history of yellowish discharge seen on one day for two consecutive months. The patient had normal secondary sexual characteristics with respect to Tanner Stage 2.

On examination, the patient's vitals were stable and her per abdominal examination was soft without muscle guarding but revealed mild tenderness over the left iliac fossa. Local examination of vulva was normal. Laboratory tests showed a total WBC count of 10,800cells/cumm with 86% Neutrophils and 13% Lymphocytes. Urinalysis was normal. CA-125 levels were reported to be 8 U/ml. Ultrasound of the patient's abdomen was done which showed left adnexa with thick walled cystic lesion measuring $5.1 \times 3.7 \times 2.7$ cm associated with partial septae and few echoes seen. CECT



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abdomen done showed both ovaries were normal with an elongated tubular peripherally enhancing hypodense structure of size $3.8 \times 5.0 \times 3.0$ cm (TR-AP-CC) noted in left adnexa with incomplete internal septations within, suggestive of hydrosalpinx. Left ovary was visualised separately from lesion (Figs. 1, 2, and 3).

Patient was posted for laparoscopy and left gangrenous tube was noted with torsion two times of the left fallopian tube along its own axis and left salpingectomy was done. Left ovary was normal. The right ovary, right fallopian tube and uterus were normal. Pathology report of necrotic tube was "consistent with Fallopian tube tissue showing extensive haemorrhage suffusion; suggestive of torsion induced changes. No atypia or malignancy". The patient tolerated the procedure well without complication. The abdominal pain subsided and the patient's post-operative recovery was uneventful and the patient was discharged 48 h later.

Discussion

A 26 year study at five paediatric surgical units conducted by Bertozzi et al. showed that only 20 cases of IFTT were reported [3]. Such cases are a rare phenomenon and are described in literature as case reports or case series. The aetiology for this diagnosis include anatomical variations such as congenital elongated mesosalpinx, hydrosalpinx, tubal or ovarian mass or malignancy [2]. Before the onset of menstruation, elevated levels of FSH may result in altered ovarian and tubal activity which may result in occlusion of the distal tube, resulting in hydrosalpinx [4]. The patient was regularly followed up for two months and had not attained menarche during the two month follow-up period. The FSH levels of the patient was checked two months after the procedure and was found to be normal at 6.91 mIU/ml. Other causes include abnormal tubal activity or venous congestion as well as trauma or pelvic surgeries with associated pelvic adhesions and Pelvic Inflammatory Disease (PID) [1]. An infectious or



Fig. 1 Ultrasound imaging of this case report showing a cystic thick wall adnexal mass





Fig. 2 Laparoscopic view showing twisting of fallopian tube twice on its own axis. Necrotic tubal tissue can be visualised adjacent to the twisted portion

pelvic inflammatory episode during childhood or uneventful torsion of the hydatid of Morgagni may increase the risk of future presentation of IFTT [4]. Studies also show that certain kinds of sports or activities that include sudden movement can result in IFTT [3]. Tubal torsion is more likely to affect the right tube considering the lack of space for torsion on the left side due to the presence of the sigmoid colon [2].

The most common presentation of adnexal torsion include lower abdominal pain that radiates to the flank or thigh and is associated with nausea and vomiting [4]. Our case was left tubal torsion with gangrenous hydrosalpinx in a 10-year old premenarchal virgo intacta with no obvious risk factors such as PID, making it a rare finding. The patient was initially sent for an ultrasound scan to rule out kidney stones based on the presenting complaints but the imaging test revealed otherwise. However the imaging test was not specific for the post-operative diagnosis of torsion of fallopian tube complicated with gangrene.

IFTT may be associated with leukocytosis and neutrophilia as is the case with our case report but is not diagnostic

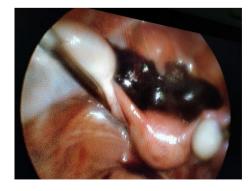


Fig. 3 Left Salpingectomy was done and image shows Laparoscopic view of the patients normal looking uterus and normal bilateral ovaries. Necrosed fallopian tube that was resected can be seen placed over the uterus

of the condition [4]. IFTT neither has a particular ultrasound finding nor particular clinical presentation; however, diagnosis is usually made during diagnostic surgery[4]. Literature reviews from previous similar cases mention that typical CT and MRI findings include thickened tube walls with cystic enlargement of adnexa [4]. However, colour Doppler may aid in identifying cases with a high likelihood of ovarian torsion by identifying presence or absence of blood flow to the twisted adnexa. It must be noted that Doppler suggestive of normal flow does not always indicate absence of torsion [4]. Majority of the cases may be diagnosed as hydrosalpinx or haematosalpinx or ovarian torsion. Other differential diagnoses to consider include appendicitis, ectopic pregnancy and ruptured ovarian cysts [1].

Salvaging the fallopian tube is quite difficult considering the fact that diagnosis is never quick or easy and it has been suggested that the risk of tubal necrosis is high if 10 h surpass the onset of pain to the surgical intervention [1]. Surgical intervention, preferably laparoscopy, is the gold standard for diagnosis and treatment for torsion of tubes by performing surgical detorsion, salpingotomy or salpingectomy [2]. Detorsion of the tube in order to return adnexal blood supply is the recommended form of conservative surgery with the purpose of increasing the chances of pregnancy in the future [2]. However, the adverse effect on future fertility such as ectopic pregnancy due to the remaining affected tube cannot be ruled out and must be explained to the patient [4].. IFTT can be managed by either laparoscopic detorsion or laparoscopic salpingectomy and both must be performed as soon as possible to avoid irreversible damage, provide faster recovery and reduce adhesions [4]. Due to the unsalvageable condition of the tube in our case, a left salpingectomy was performed.

Conclusion

In conclusion, although rare, a diagnosis of isolated tubal torsion should be considered in an acute presentation of

abdominal pain in a child or adolescent. Laboratory or imaging findings may not provide a solid diagnosis but early intervention is vital in preventing necrotic changes and adjacent ovarian gangrene. Gangrenous changes in the tube may result in severe infection and peritonitis and timely intervention will prevent severe complications and infertility. Laparoscopy is useful in diagnosis and treatment with excellent prognosis and preservation of fertility.

Declarations

Conflict of interest The authors declare that they have no conflicts of interest.

Human and Animal Rights This report does not involve animals.

Informed Consent Informed consent was taken from the patient and her parents.

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