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CASE REPORT

Ovarian Tumour Presenting as an Inguinal Hernia in a Postmenopausal Woman with Mullerian Agenesis: A case report

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ABSTRACT

Although inguinal hernias are common with inguinal hernia repairs being a common surgical procedure, ovarian inguinal hernias are rare. A 67-year-old postmenopausal woman presented with a 10-year history of right inguinal swelling on a background of primary amenorrhoea. Clinical examination revealed a 10 cm right irreducible hernia. Ultrasound and computed tomographic imaging confirmed an ovarian mass in the inguinal hernia with absent uterus and right kidney in the pelvis. Mullerian agenesis with ovarian hernia was diagnosed, however malignancy was considered due to tumour size and raised cancer antigen 125 (CA125). She underwent diagnostic laparoscopy, bilateral salpingo-oopherectomy, partial omentectomy, and inguinal hernia repair. Histopathological reports confirmed ovarian fibroma. Ovarian hernia is rare in postmenopausal women but must be considered in those presenting with inguinal masses. It can occur together with Mullerian abnormalities. CA125 can be elevated in various benign conditions, making interpretation and diagnosis difficult. Multidisciplinary approach is vital to ensure the best outcome for the patient.

Keywords: inguinal hernia, ovarian tumour, mullerian agenesis, MRKH (Mayer-Rokitansky-Kuster-Hauser) syndrome.

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Introduction

Inguinal hernia is defined as protrusion of mainly small bowels or omentum through the internal and external rings of the inguinal canal[1]. It is reported that 60% of the inguinal hernias occur on the right side, 30% on the left, and 10% bilaterally[2]. It is more common in the males compared to the females, with a ratio of 10:1. Rarely, 2.9% of inguinal hernias may contain ovaries and fallopian tubes[3]. This is mostly found in the paediatric age group and is commonly in tandem with other genital tract abnormalities[4]. Such presentation is rare in women of reproductive age group and more so in elderly women. Herein we present a case of an elderly woman with underlying Mullerian agenesis who presented with right inguinal swelling which turns out to be an incarcerated inguinal hernia with an ovarian fibroma.

Case Report

A 67-year-old nulliparous lady with underlying decompensated congestive cardiac failure, diabetes, hypertension, and thrombocytopenia, presented with a 10-year history of right inguinal swelling which was gradually increasing in size and subsequently became irreducible. She had no obvious symptoms except for occasional pain in the right inguinal region, which was worse on movement and coughing. She felt her movement had been restricted, affecting her daily activities. She also had loss of appetite and had lost 13 kg in 2 months. She denied any changes of bowel habits. Further history revealed that she never had her menses. She married late at the age of 54. However, the marriage was not consummated in the 2 years when she then lost her husband. There was no significant past surgical history.

On examination, vital signs were normal. Abdomen was soft and non-tender with a 3x3 cm umbilical hernia. There was a hard, well defined mobile mass with smooth borders measuring 9x10 cm at the right inguinal region and labia majora which was irreducible (Fig. 1). Hymen was intact with a short blind ended vagina.

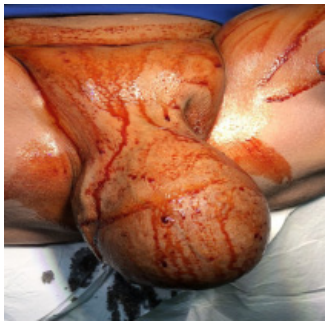

Fig. 1. Right inguinal mass

Full blood count was normal except for platelet count of 70 x 109/L. Liver function test was deranged with a total bilirubin of 30.5 µmol/L, alkaline phosphatase 235 U/L, and aspartate transferase (AST) 82 U/L. Tumour markers were taken which revealed an increased CA125 of 981 U/ml. Renal function, carcinoembryonic antigen (CEA), cancer antigen 19-9 (CA19-9), and alpha fetoprotein (AFP) levels were all normal. Ultrasound of the abdomen and pelvis was done, which revealed an absent uterus. Ovaries were not visualized clearly. The right renal fossa was empty, with the possibility of an ectopic kidney. Ultrasound of the inguinal area showed a large right inguinal mass with soft tissue and calcified component possibly a teratoma, however unable to rule out malignancy. Computed tomography scan of the abdomen and pelvis confirmed the presence of a right inguinal hernia, with a well-defined heterogenous enhancing mass

measuring 11 x 11 x 13 cm within the hernia sac (Fig. 2a and 2b). The blood supply of the mass was arising from the abdominal aorta. These features, along with the raised CA 125, suggested a likely ovarian tumour. Subcentimetre para-aortic and paracaval nodes were present, but there was no ascites. The left ovary was normal, but normal uterus was not seen. The left kidney was normally located, whereas the right kidney was seen in the right pelvic region. There was a small right paraumbilical hernia containing omental fat. There was also liver cirrhosis with splenomegaly and gastro-esophageal varices. A diagnosis of suspected Mullerian agenesis with paraumbilical and ovarian hernia unable to exclude malignant transformation was made. Referral was made to the gastroenterology team for further management of liver cirrhosis.

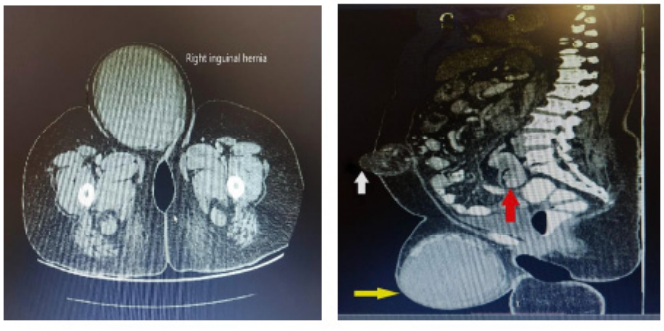

Fig. 2a (left): Axial view of computerized tomography (CT) scan showing right inguinal hernia. **Fig. 2b** (right): Sagittal view of CT scan showing right inguinal hernia containing ovarian tumour (yellow arrow) and umbilical hernia (white arrow); right ectopic kidney noted (red arrow)

The patient underwent diagnostic laparoscopy, bilateral salpingo-oophorectomy, partial omentectomy, and inguinal hernia repair. The surgery was performed together with the surgical team. Intraoperatively, diagnostic laparoscopy revealed absent right rudimentary uterine bud. Right ovary, infundibulopelvic ligament and vessels, as well as omentum were found herniated through the right inguinal canal. The left uterine bud was present with normal left fallopian tube and ovary. There was no ascites and no suspicious tumour nodules seen. Left salpingo-oopherectomy was done laparoscopically. From below, the right inguinal mass was excised (Fig. 3a). The skin defect was refashioned. The skin was closed interruptedly with a drain (Fig. 3b). The patient recovered uneventfully. The histology was consistent with right ovarian fibroma, a benign ovarian tumour (Fig. 4).

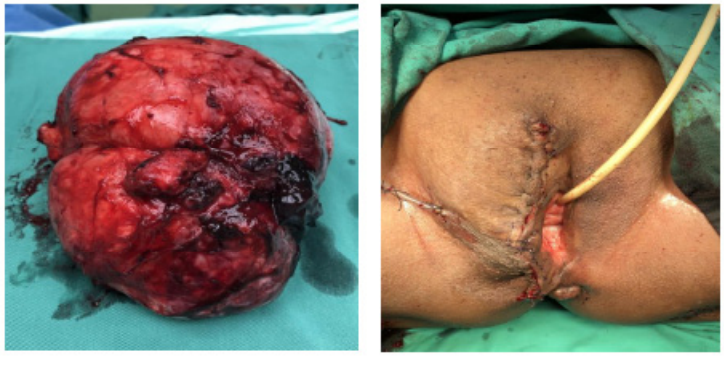

Fig. 3a (left): Right ovarian tumour. **Fig 3b** (right): Skin refashioned and closed interrupted with drain in situ.

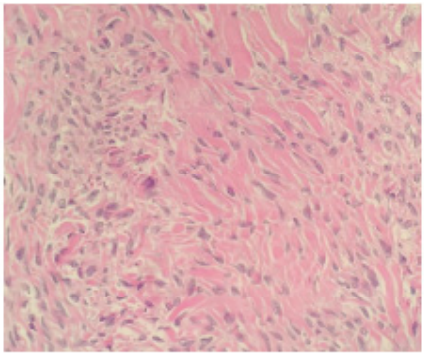

Fig. 4. Spindle shaped cells arranged in fascicles and haphazard pattern. No cellular atypia, necrosis or mitosis

Discussion

The case presented is unique due to the multiple factors which made diagnosis challenging. Firstly, our patient was unfortunate enough to have double pathology, which is the Mullerian agenesis and an ovarian tumour manifesting as an inguinal hernia. The common link between Mullerian abnormalities and ovarian hernias is the embryogenic structure called gubernaculum. Paired gubernaculum attaches to the caudal part of the gonads and helps to guide the descent of the gonads to their proper place in the developing fetus. In females, the upper part of the gubernaculum, together with the ovarian artery and vein, becomes the suspensory ligament of the ovary which helps to suspend the ovary to the pelvic side wall. The lower part of the gubernaculum gives rise to the round ligament and the ovarian ligament. The round ligament then courses through the inguinal canal and attaches to the labia majora (Fig. 5). Gubernaculum dysfunction seems to be related to pathologies arising from the round ligament and inguinal hernia, as well as to Mullerian duct abnormalities[5].

Furthermore, it is suspected that there is underlying weakness of the ligaments that hold the ovaries in place to result in herniation[6]. Weakening and lengthening of the supportive ligaments are exacerbated in multiparous women and those with conditions that result in frequently increased intra-abdominal pressures, such as chronic cough[6]. There is also hypothesis that failure of fusion of the Mullerian ducts lead to excessive mobility of the ovaries and the uterine cornua, making it more likely for these structures to herniate through the inguinal canal[7], as in the case of our patient.

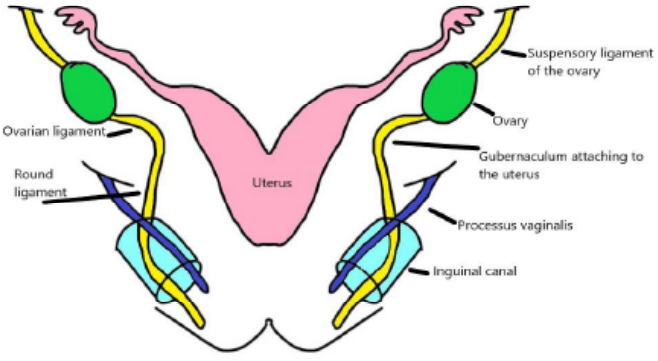

Fig. 5. Gubernaculum and its derivatives

Ovarian fibromas are solid tumours belonging to sex cord stromal cell tumours of the ovary, comprising of spindle shaped fibroblastic cells and abundant collagen[8]. They are the most common benign solid tumours of the ovary, occurring mainly in women in the 5th decade of life. In a small number of cases, ovarian fibromas can also result in raised CA 125 levels[8].

It is well known that raised CA 125 is not diagnostic of ovarian malignancy. It can be raised in many benign gynaecological and non-gynaecological conditions as well. Cirrhosis of the liver is one of the most common disorders associated with increased levels of CA125, occurring in 85% of cases[9]. This could explain the reason behind the raised CA 125 as well as the thrombocytopenia. Many patients with cirrhosis also experience fatigue, anorexia and weight loss[10], mimicking constitutional symptoms of malignancy, as demonstrated with our patient. This highlights the complexity of interpretating CA 125 in the presence of liver disease.

There have been other reported unusual contents discovered in inguinal hernias. Shetty et al[11] report a case of a 66-year-old who presented with a 3-year history of a slowly enlarging right inguinal mass. Ultrasound and computed tomography scan were done prior to surgery. Intraoperatively, the right inguinal hernia was found to contain omentum, caecum, and an ovarian cyst, which turned out to be a mature cystic teratoma. Other atypical inguinal hernia contents include uterus[12] and even endometriosis[13,14], although the latter is mainly found in the premenopausal age group.

One must always consider the possibility of malignancy in such cases too. Although extremely rare, there have been reported cases of ovarian malignancy presenting as inguinal ovarian hernia. Burke et al[15] describe an 89-year-old lady presenting with bilateral inguinal hernias diagnosed to have metastatic ovarian carcinoma and was managed conservatively due to extensive disease and advanced age. Hung et al[16] report a 48-year-old woman with stage 4 ovarian carcinoma with carcinomatosis presenting initially as bilateral inguinal hernias. In this case, the diagnosis was only made after histopathological examination of the excised hernial sac revealed an adenocarcinoma with unknown primary. Further investigations revealed the ultimate diagnosis of ovarian carcinoma and patient responded well to chemotherapy. This highlights the importance of imaging when dealing with inguinal masses. Most of the time, the diagnosis of inguinal hernia is made clinically. However, as these cases have demonstrated, there must be a high index of suspicion as to what the inguinal hernia might contain and if possible, efforts made to identify the contents before surgery. In our case, possible malignancy was a reasonable preliminary diagnosis to make given her age, size of tumour, presence of constitutional symptoms, and markedly raised CA 125 levels.

Conclusion

Ovarian hernia is rare in postmenopausal women but must be considered in those presenting with inguinal masses. It can occur together with Mullerian abnormalities. CA125 can be elevated in various benign conditions, making interpretation and diagnosis difficult. Multidisciplinary approach is vital to ensure the best outcome for the patient.

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Potential conflicts of interest

The authors declare no conflicts of interest.

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