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Pure Sertoli Cell Ovarian Tumour: a rare cause of postmenopausal adnexal torsion

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Introduction

This report describes cumulatively rare findings in a postmenopausal woman (PMW) with left sided adnexal torsion involving a Pure Sertoli Cell tumour.

Case Report

A 62-year-old P3 PMW, presented with a 5-hour history of progressive left sided acute abdominal pain. She had generalised abdominal tenderness with no peritonism and a large pelvic mass ~16 weeks' size. There were no other abnormal clinical findings.

CT demonstrated a large left-sided mixed cystic/solid mass extending into the right abdomen, likely ovarian. The left pedicle appeared twisted with fluid in the pelvic and left paracolic gutter suggesting ovarian torsion.

Laparotomy revealed a large (23cm x 18.5cm x 14cm) ovarian mass twisted around the left suspensory ligament thrice (see Figure 1). The mass was smooth and lobulated, with areas of haemorrhage (see Figure 2) and a left salpingo-oophorectomy ensued. The right ovary was normal in appearance. Recovery was uneventful. Histopathological findings demonstrated Sertoli cells with no Leydig cells. IHC stained positive for calretinin, confirming diagnosis of Sertoli cell tumour (FIGO stage IA). CA-125 and testosterone were normal.

Discussion

Adnexal torsion, the twisting of the suspensory ligament supplying the ovary resulting in ischaemia, is rare in PMW and requires a high index of suspicion^[1].

Typically presenting with abdominal pain, nausea, and vomiting, it is a known gynaecological emergency^[2]. Unlike in this case, where only a left salpingo-oophorectomy was performed, management is usually more radical in PMW, involving bilateral salpingo-oophorectomy with or without hysterectomy^[1].

Whilst histological findings most commonly reveals a benign ovarian tumour, the risk of malignancy, although not unheard of, is rare, accounting for ~3%^[1] ^[2]. The finding in this case was that of a Sertoli cell tumour (SCT), an uncommon variant of the Sertoli-Leydig cell tumours (SLCTs) of which less than (<)10% present in PMW^[4]. SLCTs are an extremely rare type of sex cord stromal tumour (SCST) of the ovary which account for less than (<) 1% of all SLCTs and <0.5% of all primary neoplasms^[4]. SLCTs typically localize to the right, unlike this case which involved the left ovary. Pure Sertoli cell tumours are an even more rare variant of SLCTs, accounting for ~ 4% and, by definition, lacks Leydig cells in the stroma^[4], as was the case in this patient. The characteristic presentation is mainly endocrine, with virilization being predominant but not obligatory. This patient had normal testosterone levels and no discerning androgenic features.

Serum CA-125 level can be elevated in sex cord ovarian tumours including pure SCT and studies have shown that pre-operative serum CA-125 determination has a clinical value in predicting the curability and stage of the disease ^[4]. This patient's CA-125 was normal.

Most patients with SLCTs (81.8% - 100%) have stage 1 cancers according to the International Federation of Gynaecology and Obstetrics (FIGO)^[4], as was the finding in this patient. Good prognostic factors are low grade, low stage and absence of heterologous elements ^[4]. The overall 5-year survival rate was reported as 95% for stage 1 disease^[3]. Given the early-stage disease in this patient, a good prognosis is expected.

Conclusion

Adnexal torsion is a gynaecological emergency and presentation in PMW is rare, requiring a high index of suspicion. Bilateral salpingo-oophorectomy is the recommended management of choice. The diagnosis of an ovarian malignancy in the PMW is rare, <3% and rarer still is the finding of a pure SCT. Prognosis is generally favourable for early-stage disease.

References

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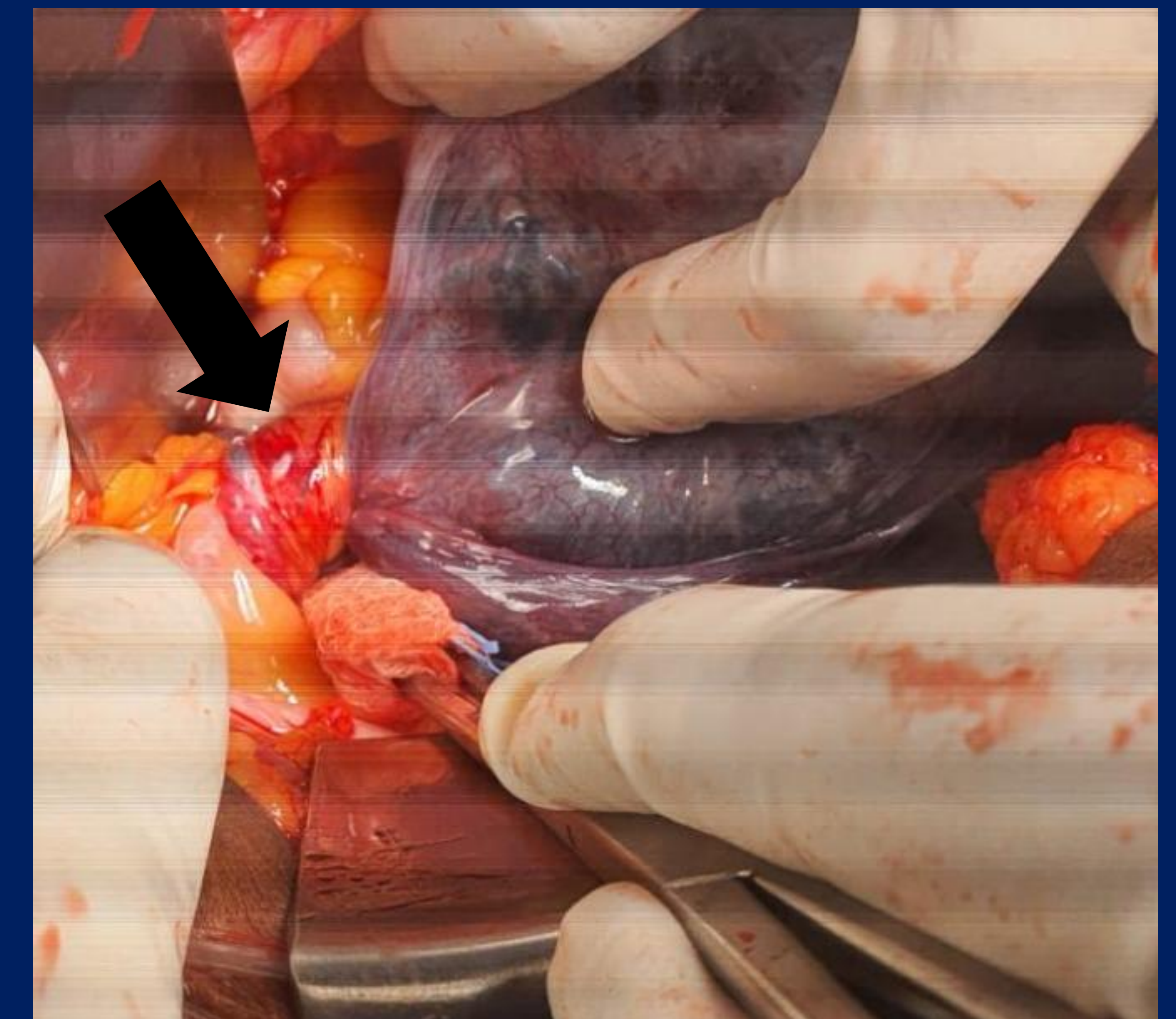


Figure 1: Torted infundibulo-pelvic ligament indicated by the black arrow.

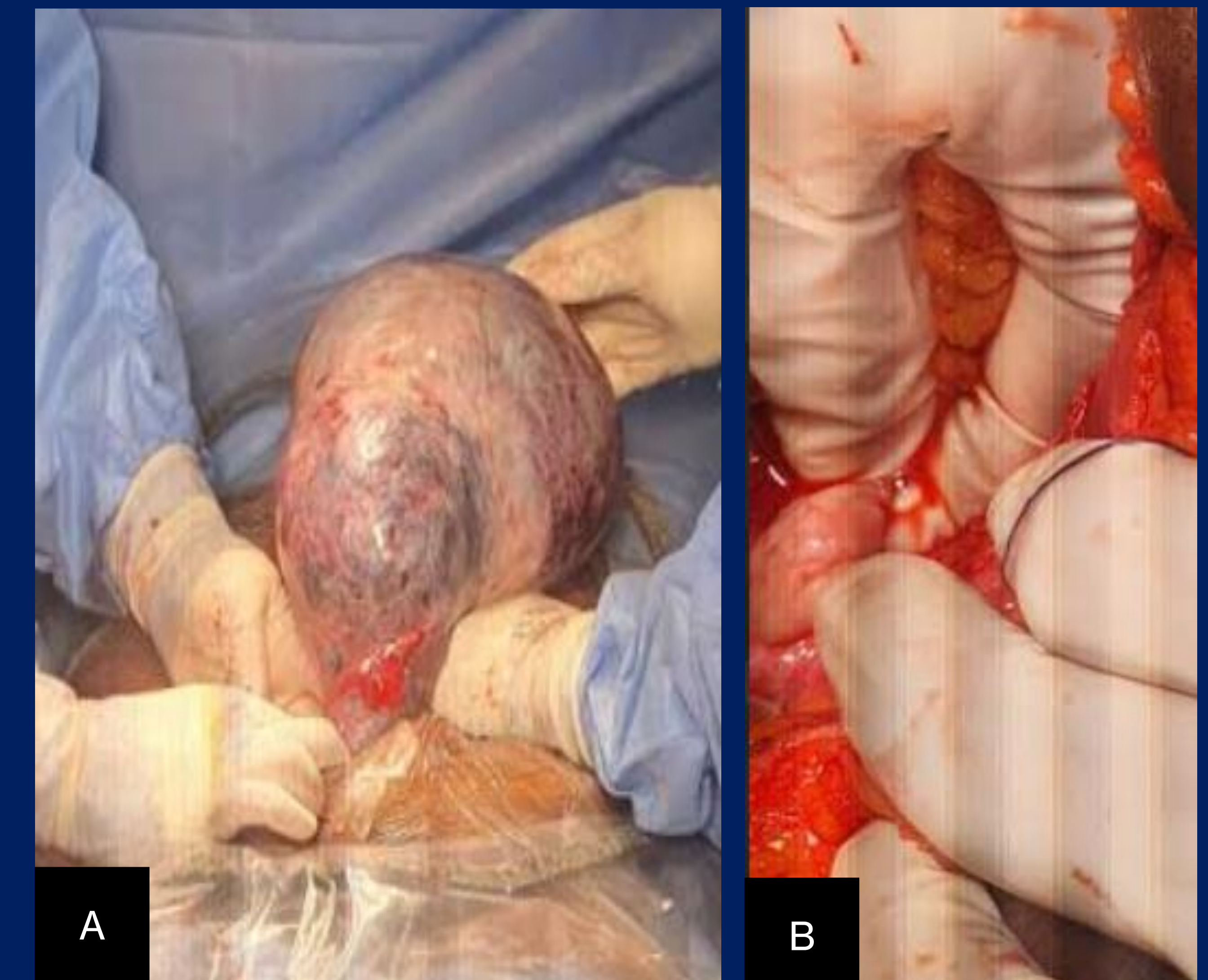


Figure 2: A) large left ovarian mass and B) normal right ovary