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Ovarian Remnant Syndrome. A Case Report with Literature Review

C. Spyroulis[1*]
N. Matthews[2]
S. Al-Inizi[3]

[1]Specialty trainee in Obstetrics and Gynaecology, South Tyneside and Sunderland NHS Foundation Trust, UK
[2]Consultant Gynaecologist and Obstetrician, South Tyneside and Sunderland NHS Foundation Trust, UK
[3]Consultant Gynaecology and Obstetrician, South Tyneside and Sunderland NHS Foundation Trust, UK

Abstract

Ovarian remnant syndrome (ORS) is a rare and very challenging clinical condition where small pieces of ovarian tissue are inadvertently left in the pelvic cavity following the surgical removal of one or both ovaries. The remnant tissue can grow forming cysts causing pelvic pain [1]. it was initially described by Shemwell and Weed in 1970 [2]. Pelvic pain, cyclical or chronic, is a common symptom. Other symptoms can include dyspareunia, dysuria and other urinary and bowel symptoms [3,4].

Management of ORS is challenging especially it is commonly associated with adherent cystic structures to vital organs such as the ureter, bladder or bowel [1]. Due to its complex nature, it should be approached and managed at a multidisciplinary (MDT) level. We are presenting a case of ORS in a 39-year-old lady following laparoscopic bilateral oophorectomy for endometriosis. Her right ovary was densely adherent to the right ureter resulting in a very small ovarian tissue left behind adherent to the ureteric wall. This resulted in recurrence of chronic pelvic pain and regrowth of a right ovarian mass which was managed by a multidisciplinary team (MDT).

Article Highlights:

- Ovarian remnant syndrome (ORS) is defined by the presence of a pelvic lesion related to residual ovarian tissue after oophorectomy.
- It is a rare condition, but extremely challenging to diagnose and treat.
- Pelvic pain either cyclical or most commonly chronic, is a common symptom.
- Multidisciplinary input is required to manage ORS.

Case Report

A 39-year-old lady who had two previous normal deliveries and known to have endometriosis, presented with recurrence of chronic pelvic pain following laparoscopic bilateral salpingoopherectomy and an ovarian complex mass was identified on imaging.

Her previous gynaecological history dated back to 2011 when she had vaginal hysterectomy at South Tyneside Hospital, South Shields, UK, due to persistent CIN 3 (cervical intra epithelial neoplasia) following multiple loop excisions of her cervix. She presented in 2014 with increasing lower abdominal pain which was severe enough to necessitate admission and an ultrasound scan (US) of the pelvis confirmed large elongated tubular right sided (8x3.3 cm) cystic structure suggesting a right hydrosalpinx. She had emergency laparoscopy which indicated a large tortuous hydrosalpynx occupying most of the pelvis adherent to the pelvic wall with evidence of active endometriosis. Both ovaries were adherent to each other and to the top of the vagina with bowel adhesions. Adhesions were divided and laparoscopic bilateral salpingectomy was performed uneventfully. She continued to have chronic pelvic pain and a follow up ultrasound scan in 2015 showed a small 3.6 cm right ovarian cyst. She was started on Zoladex 3.6mg (Goserelin Acetate, AstraZeneca UK Limited) once per month for 6 months. This had improved her pelvic pain. The patient requested to have her right ovary removed. Laparoscopy indicated that both ovaries were of normal size and were attached to each other on the top of the vaginal vault and were both adherent to ureters, more on the right side with an active endometriotic nodule on the right iliac vessel and right ovarian fossa. Decision was not to perform right oophorectomy without urological imaging especially the patient would benefit from bilateral oophorectomy for active endometriosis after counselling regarding operative risks and surgical menopause. Intravenous urography was later performed indicating intact ureters and a urological input was sought. A joint gynaecological/urological procedure performing laparoscopic bilateral oophecretomy using LigaSure Maryland (Jaw Open and Laparoscopic Sealer/Divider with Nano-coating Technology, Medtronic, UK) and ureteric stenting was undertaken in 2016. The right ovary was densely adherent to the right ureteric wall, a small remnant of 2 mm

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*Corresponding author:

Christos Spyroulis

Sunderland Hospital
Department of Obstetrics and Gynaecology
Sunderland, SR4 7TP
Tel: +447570543874
Email: christosspyroulis@doctors.org.uk

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Figure 1: Image before the removal of the ovary

Figure 2: Image after the removal of ovary

ovarian tissue was left in situ as advised by the urologist to avoid right ureteric fistula. The patient had a smooth post-operative recovery and was pain free 3 months post-surgery. Histology confirmed that both ovaries were removed (slightly smaller right ovary-35x25x25mm & 30x25x20 mm) with no malignancy identified. Two years later, she presented with recurrence of chronic pelvic pain and an US scan and CT urogram showed a 3.5cm midline complex cystic lesion in the rectovaginal space without a visible distant plane between the cyst and the anterior wall of the mid-rectum with a visible plane of separation from both ureters. She was restarted on Zoladex injections (3.6 mg, Goserelin Acetate, AstraZeneca UK Limited) which improved her pain. The case was discussed at the urology MDT meeting and removal of the ovarian remnant tissue was planned as a joint procedure with the gynaecologist which could require ureteric resection and end to end anastomosis. Laparoscopy indicated a right sided cystic lesion attached to vaginal vault and sigmoid colon with close proximity to the bladder and right ureter which was stented. The lesion was removed without jeopardising the bladder and right ureter and was sent for histology. Methylene blue was injected in the bladder which ruled out any injury. Histology indicated ovarian tissue 43x30x20 mm with multiple haemorrhagic cysts and no evidence of malignancy. She recovered well and was discharged back to the care of her GP (Figure 1 and 2).

Discussion

ORS is the presence of ovarian tissue in a patient who has had previous oophorectomy which has to be confirmed histologically [1]. This definition has been broadened to include women who had unilateral oophorectomy with remaining ovarian tissue on the ipsilateral side [5]. It presents with pelvic pain or pelvic mass.

Generally, it is a benign condition, with some might have a malignant potential [6]. Atypical endometriosis of the ovary appears to represent a transition from benign endometriosis to carcinoma [7]. ORS can rarely be presented in different sites such as at a laparoscopy port [8].

The main risk factors for ORS are endometriosis, pelvic inflammatory disease/tubal disease, increased pelvic vascularity, pelvic adhesions and altered anatomy [9]. It was previously reported that ORS develops more frequently on the left pelvic side wall [10], however, in this case it was on the right.

ORS is a rare condition which was reported in around 1%-3% of post hysterectomy and bilateral oopherectomy cases [11]. There is evidence in the literature to support that devascularised tissue can re-implant on intact or denuded peritoneal surfaces and the re-vascularised tissue can become functional as evidenced by follicle formation and hormonal production [1,12]. For the diagnosis of ORS, an ultrasound scan of the pelvis can be used especially when there is a pelvic cyst/mass, but also MRI in more difficult cases can be helpful. Checking hormones such as FSH, LH and estradiol levels can help to make the diagnosis [13]. In very difficult cases to localise ovarian tissue before resection; a course of ovulation induction with clomiphene citrate (clomid 50 mg, William s. Merrell, UK) can be utilised [14]. In the literature, the therapeutic options of ORS are medical and pelvic irradiation therapy to obliterate the ovarian remnant tissue if surgery is difficult or the patient declines surgery [15,16].

We describe a case of ORS where the right ovary was densely adherent to the right ureteric wall and complete removal of that ovary would have resulted into ureteric fistula, hence a very small remnant was left in situ. She unfortunately returned with recurrent pelvic pain and an ovarian mass necessitating hormonal therapy followed by surgical excision with an MDT involvement due to its challenging nature involving different organs. The case highlights the importance of counselling patients with risk factors for ORS such as endometriosis about the possibility of the syndrome despite it is rare. The condition can be prevented by retroperitoneal dissection to facilitate high ligation of the infundibulopelvic ligament with excision of all peritoneum and tissue adherent to the ovary [17]. This was impossible in the case due to the severe adherence of the ovary to the ureteric wall especially the patient was not consented for ureteric resection and implantation.

Conclusion

ORS is a rare and very challenging condition. It is one of the causes of post hysterectomy and oophorectomy pain and pelvic mass formation especially in women with endometriosis. Management needs careful imaging and MDT input to facilitate surgical resection.

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References

1. Webb MJ. Ovarian remnant syndrome. Aust N Z J Obstet Gynaecol. 1989 Nov;29(4):433-435.
2. Shemwell RE, Weed JC. Ovarian remnant syndrome. Obstet Gynecol. 1970;36:299.
3. Magtibay PM, Magrina JF. Ovarian remnant syndrome. Clin Obstet Gynecol. 2006 Sep;49(3):526-534.
4. Kooperman RS. Diagnosis, treatment and prevention of ovarian remnant syndrome. Ob Gyn News. 2019 Jul.
5. Arden D, Lee T. Laparoscopic excision of ovarian remnants: retrospective cohort study with long-term follow-up. J Minim Invasive Gynecol. Mar-Apr 2011;18(2):194-199.
6. Kho RM, Abrao MS. Ovarian remnant syndrome: aetiology, diagnosis, treatment and impact of endometriosis. Curr Opin Obstet Gynecol. 2012 Aug;24(4):210-214.
7. Munkgaard PS, Blarkaaer J. The association between endometriosis and ovarian cancer: a review of histological, genetic and molecular alterations. Gynecol Oncol. 2012 Jan;124(1):164-169.

8. Tilstra M, Vaught JM. Ovarian Remnant Syndrome in Ovarian Tissue Ectopically Implanted in Trocar Site: A Case Report. J Min Invasive Surg. 2016;23(7) Supplement, S86.
9. Chao HA. Ovarian remnant syndrome at the port side. J Minimal invasive Gynecol 2008;15:505-507.
10. Nezhat F. Nezhat C. Operative laparoscopy for the treatment of ovarian remnant syndrome. Fertility and Sterility. 1992 May;57:1003-1007.
11. Christ JE, Lotze EC: The residual ovarian syndrome. Obstetrics and Gynaecology 1975 Nov;46:551-556.
12. Minke T, DePond W, Wimkelmann T, et al: Ovarian remnant syndrome: Study in laboratory rats. Am J Obstet Gynecol. 1994 Dec;171(6):1440-1442.
13. Price FV, Edwards R, Buchsbaum HJ: Ovarian remnant syndrome: Difficulties in diagnosis and management. Obstet Gynecol Surv. 1990 Mar;45(3):151-156.
14. Orford VP, Kuhn RJ. Laparoscopic resection of ovarian remnant after clomiphene-assisted ultrasound localization. Gynaecol Endosc 1997 Feb;6(1):55–56.
15. Richlin SS, Rock JA: Ovarian remnant syndrome. Gynaecol Endosc. 2002 Jul;10:111-117.
16. Abu-Rafeh B, Vilos GA, Misra M. Frequency and Laparoscopic Management of Ovarian Remnant Syndrome. J Am Assoc Gynecol Laparosc. 2003 Feb;10(1):33-37.
17. Wassn M, Magrina J. Surgical management and prevention of Ovarian Remnant. J Minim Invasive Gynecol. Jul-Aug 2019;26(5):811.