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Case Report

Coexistence of Struma Ovarii and Endometriomas - A Case Report and Review of the Literature

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Abstract

Struma ovarii is a rare, monodermal variant of mature ovarian teratoma predominately composed of mature thyroid tissue. It is usually diagnosed on histopathology following removal of an adnexal mass. Co-occurrence of endometriosis and Struma ovarii has been described in two case reports with no cases described of this entity co-existing with endometrioma in the same ovary in the absence of a dermoid cyst. A 47-year-old patient presented with a history of Stage IV endometriosis. Following laparoscopic oophoro-cystectomy, on histopathology assessment, Struma ovarii was diagnosed within an endometrioma. The patient had a normal TSH and CA-125. She recovered well post operatively and is seeking further fertility treatments using donor oocytes. This case presents the unique finding of Struma ovarii co-existing with endometrioma in the same ovary.

INTRODUCTION

Endometriosis is the presence of functional endometrial glands and stroma outside of the endometrial cavity. It can present with dyspareunia, dysmenorrhea, chronic pelvic pain and/or infertility [1-3]. Endometriosis can be diagnosed intraoperatively by visualization and confirmed with histopathology [2]. General prevalence ranges between 10 and 20% but is estimated to be as high as 50% in women with infertility, and 65% in women with chronic pelvic pain [2].

Struma ovarii is a rare monodermal type of mature ovarian teratoma, predominately composed of mature thyroid tissue, originating from the primitive germ cell layer [4-6]. It is generally diagnosed incidentally on histopathology following excision of a unilateral, solid-cystic pelvic mass and comprises 0.3-1% of solid ovarian masses [4,5,7]. Diagnosis requires that at least 50% of the mass be composed of thyroid tissue [5,7-9]. The majority are diagnosed after age 40 and the treatment is surgical removal [4,5]. In approximately 5% of cases, signs and symptoms of hyperthyroidism have been reported [5,7,10]. Struma ovarii can be malignant in 5 to 10% of cases [4,5,8]. Although the CA-125 is usually found to be normal, cases of elevated levels have been reported [8,9].

Several cases of coexisting endometrioma and mature cystic teratoma have been described [3,4]. Campo et al., reported a case where metastatic lesions of recurrent Struma ovarii were found at the time of surgery for an ovarian endometrioma [7]. To the best of our knowledge, this is the first report of an incidental finding of Struma ovarii coexisting with an endometrioma in the same ovary. This case emphasizes the importance of resection of a complex adnexal cyst in its entirety.

CASE PRESENTATION

This case report is from The Fertility Clinic in London, Ontario Canada. Institutional Review Board review was waived and signed consent was obtained from the patient. A 43-year old woman initially presented with a history of secondary infertility of ten months' duration. Her history was significant for Crohn's disease and mild hypothyroidism, with a baseline TSH of 4.37 mIU/L. She had never undergone any surgeries. She was previously found to have had a 2.3 x 3.4 cm cyst, query a hydrosalpinx. On presentation, a spontaneous pregnancy was diagnosed, which unfortunately resulted in a spontaneous abortion.

An ultrasound performed in follow up of the patient's miscarriage showed a cyst on the right ovary, 5.5 cm x 3.7 cm in size, with mild internal echoes suggestive of blood. The patient then underwent multiple ovulation induction cycles. Seven months later, during an ovulation induction cycle, she presented with a several week history of left lower quadrant abdominal pain. A trans-vaginal ultrasound at that point showed bilateral ovarian cysts. On the right side, a 5.1 cm x 3.2 cm cyst was noted with a septum running through the middle. On the left ovary, a 6.3 cm by 5 cm cyst was visualized with a hyperechoic component and a septum. Surgical intervention was therefore planned.

The patient underwent laparoscopic lysis of extensive tubal adhesions bilaterally and partial excision of a left ovarian cyst. In addition, two ovarian endometriomas were drained from the left ovary, chromopertubation indicated bilateral tubal occlusion, and a left para-ovarian cyst was removed. Histopathology examination of the left ovarian cyst revealed the presence of fibrous tissue with no epithelial lining or ovarian tissue identified. The para-ovarian cyst was a benign mesothelial cyst on histopathology. Post-operatively, the patient elected to trial treatment with dienogest, 2 mg daily by mouth (Visanne, Bayer, Mississauga, ON) for suppression of her endometriosis. Following this, she sought further fertility treatment at a different clinic, and was lost to follow-up.

Three years after the initial presentation, at 47 years of age, the patient was again seen at our centre in the context of undergoing remote ultrasound monitoring during fertility treatment. She reported a significant improvement in her pelvic pain symptoms. Sonography showed a simple cyst on the right ovary, measuring 2.7 cm and a simple cyst on the left ovary, measuring 9.3 x 7.3 cm, with a septated region. There was no free fluid seen in the pelvis. CA-125 was normal at 15 U/mL and a plan was made for repeat ultrasound in six to eight weeks' time. A laparoscopic oophoro-cystectomy was discussed with the patient at this point. Within two weeks, she presented to the Emergency Department of our hospital with left lower quadrant abdominal pain with concerns of an ovarian torsion. An ultrasound showed the 9.5 cm cyst in the left ovary with low level internal echoes, a normal arterial waveform and a difficult to identify venous waveform. A 2.4 x 2.3 x 2.0 cm cyst was again seen in the right ovary with low-level internal echoes. There was no free fluid seen and she was treated conservatively for pain control followed by a gonadotropin releasing hormone (GnRH) antagonist (Elagolix, Abbvie, St. Laurent, QC) for worsening of pelvic pain and bowel irritation symptoms. During follow up, the patient was scheduled for an MRI, which showed a left ovarian hematosalpinx-endometrial complex with the left endometrioma measuring up to 10.0 cm (Figure 1) with a 0.8 cm T2 dark spot along the lateral wall of the left ovarian lesion and a thin septation. In addition, note was made of a suspected right ovarian hemorrhagic cyst and uterine adenomyosis. There was no pelvic free fluid or lymphadenopathy noted. Laparoscopic surgery was planned.

RESULT

In the operating room, hysteroscopic examination was normal. At laparoscopy, a large tubo-ovarian mass was seen on the left side with the sigmoid and epiploica being adherent to the mass. The uterus appeared normal and the right ovary and tube were not visualized as they were engulfed in omental and peritoneal adhesions in the cul-de-sac. Using the CO2 laser, first adhesiolysis was performed of the left cyst followed by draining serous fluid with some old clot at the base of the cyst consistent with old endometrioma fluid and subsequent oophoro-cystectomy. The post-operative diagnosis was that of Stage IV endometriosis with extensive pelvic and cul-de-sac adhesions, a left serous ovarian cyst and old endometrioma.

The histopathology report described the left ovarian cyst to be a hemorrhagic cyst suggestive of endometrioma. This was based on the presence of focal endometrial stroma and cuboidal type epithelium, as well as hemosiderin laden macrophages combined with the clinical history of stage IV endometriosis (Figure 2). In addition, note was made of a small lesion consisting of thyroid follicles, reported as Struma ovarii. Thyroglobin and Thyroid Transcription Factor-1 immunostaining were both positive at the Struma ovarii component, confirming thyroid origin (Figure 2). Of note, the lining of the cyst was negative for Thyroid Transcription Factor-1 immunostaining (Figure 2).

Post-operatively, the TSH was normal at 1.79 mIU/L. The patient recovered well after surgery and is continuing with fertility treatment using donor oocytes.

DISCUSSION

Struma ovarii is a monodermal mature teratoma composed of at least 50% thyroid tissue [4-6]. It is rare, representing only 1% of ovarian tumours and 2.7% of dermoid tumours [9]. Since it usually presents with an asymptomatic pelvic mass with non-specific findings on imaging, it is often only diagnosed post-operatively following histopathologic evaluation [4]. This tumour type can be found at any patient age, but is usually diagnosed in the fifth and sixth decades of life [7]. Although usually benign, these tumours have been found to be malignant in 5-10% of cases and should be treated by surgical excision [4,5,8]. Endometriosis, on the other hand, is a relatively common disorder frequently seen in women with chronic pelvic pain and/or infertility [2]. In some women, it can cause severe and debilitating symptoms including dysmenorrhea, dyspareunia, and non-cyclical chronic pelvic pain [1].

A unique case described by Campo et al. was that of a 20-year-old female who had prior surgical treatment of Struma ovarii at 12 years of age. The patient then presented with an adnexal mass eight years after the initial surgery and was found to have an ovarian endometrioma and multiple Struma ovarii implants throughout her peritoneal cavity concerning for the follicular variant of papillary thyroid carcinoma on histopathology [7]. Hwang et al. also reported a case in a 22-year-old woman who underwent surgery for bilateral dermoid cysts as well as a left endometrioma. She was found to have Struma ovarii on histopathology in addition to the mature cystic teratoma and endometrioma in the left ovary [4].

Our case is unique in that Struma ovarii was found incidentally in an ovary with endometrioma in the absence of any evidence of a coexistent dermoid cyst. This may represent an early presentation of Struma ovarii. Our case emphasizes the importance of careful resection of entire pelvic cyst contents followed by detailed evaluation and confirmation of the diagnosis with histopathology.

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