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Case Report

Extra-Lymph Node Retroperitoneal Recurrence of a Serous Borderline Ovarian Tumor

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Abstract

We report the case of a 32-year-old woman presenting a serous Borderline Ovarian Tumor (BOT) in the retroperitoneal space revealing ovarian and para-aortic lymph node recurrence. The patient had been treated previously by unilateral salpingo-oophorectomy for a serous BOT with non-invasive peritoneal implants. Extra-lymph node retroperitoneal recurrence of serous BOT has previously been reported in two patients and the physiopathology of such recurrence remains to be determined. Serous BOT can exhibit unusual late distant recurrence.

Keywords: Lymph node recurrence; Micropapillary patterns; Ovarian carcinoma; Retroperitoneal space; Stromal microinvasion

Introduction

Borderline Ovarian Tumors (BOT) differ from ovarian carcinoma by the absence of stromal invasion [1]. While the true incidence is unknown, around 10% of malignant ovarian tumors are estimated to be BOT [1,2]. Data from the Surveillance Epidemiology and End Results (SEER) program showed an incidence of 2.5 per 100 000 women-years in the USA [3]. About 15-20% of serous tumors are borderline [2] representing the most frequent BOT [4]. However, a Danish register-based cohort study found 50% of BOT to be mucinous tumors suggesting variations in incidence according to ethnicity and country [5]. Serous BOT are bilateral in 15-40% of cases and between 15-40% are also associated with extra-ovarian disease (peritoneal implants or nodal disease) [2]. In 1996, the concept of serous BOT displaying micropapillary patterns was introduced [6]. These lesions are characterized not only by specific morphological criteria but also by a propensity for invasive peritoneal implants and lymph node involvement. However, extra-lymph node retroperitoneal location or recurrence is rare [7]. We report a patient presenting a serous BOT in the retroperitoneal space revealing ovarian and para-aortic lymph node recurrence.

Case Report

In October 2017, a 32-year-old woman was referred to the Department of Urology for right lumbar pain. MRI showed a retroperitoneal cystic lesion centered on the neurovascular pedicle of the 12th right rib with a cystic component in contact with the posterior surface of the kidney, and a para-aortic lymph node (Figure 1). The retroperitoneal mass was resected in November 2017, and the neurovascular pedicle of the 12th rib, the 12th rib and the renal fascia were removed. Histopathology revealed a papillary and cystic serous tumor of tubo-ovarian origin (PAX8 + profile, RE+ / RP+, WT1 +, P16 +) with a low proliferative index suggesting a retroperitoneal location of a serous BOT or a metastatic localization of a low-grade serous carcinoma. The renal fascia showed two superficial non-invasive micro-foci of a papillary tumor (Figure 2).

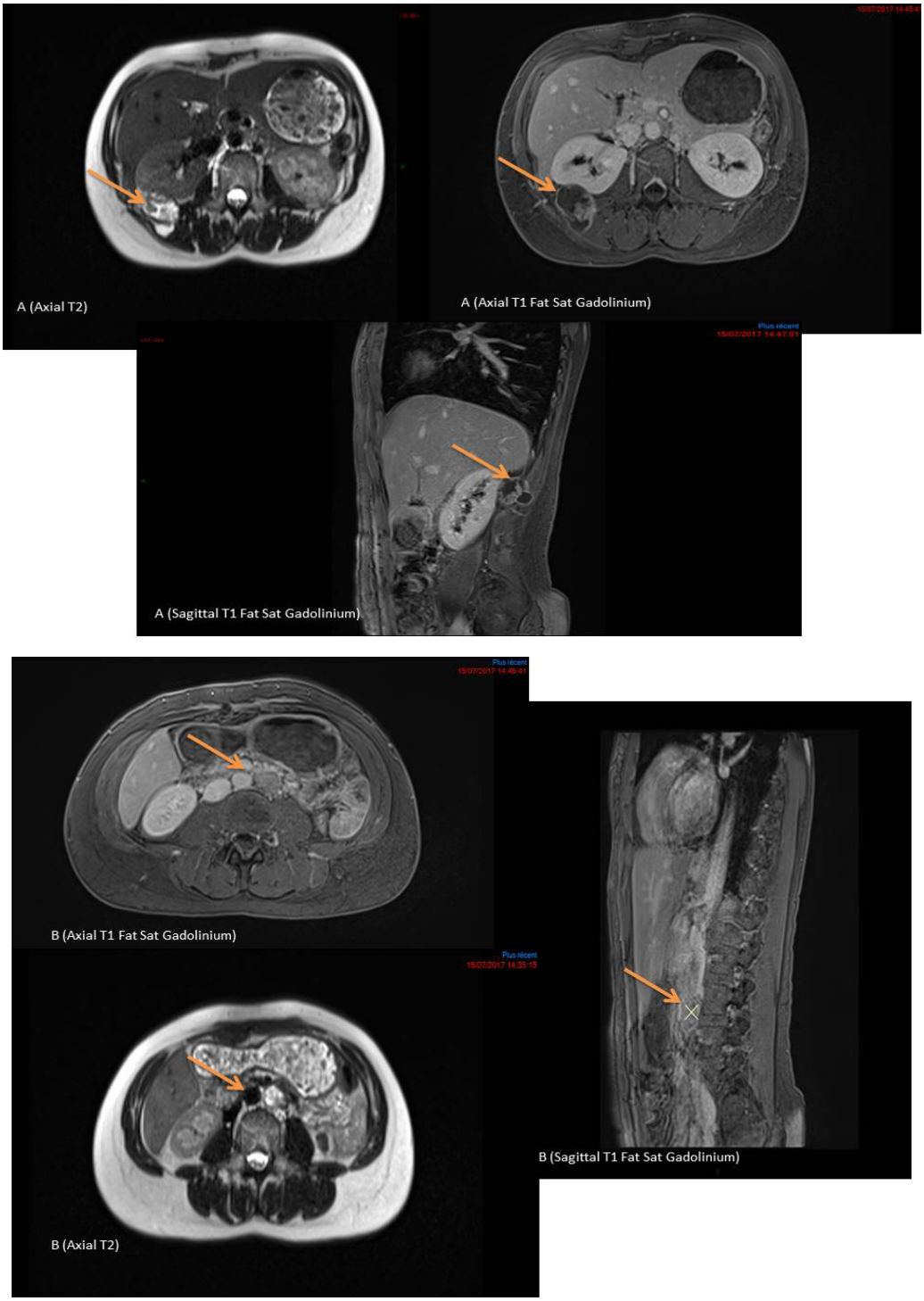
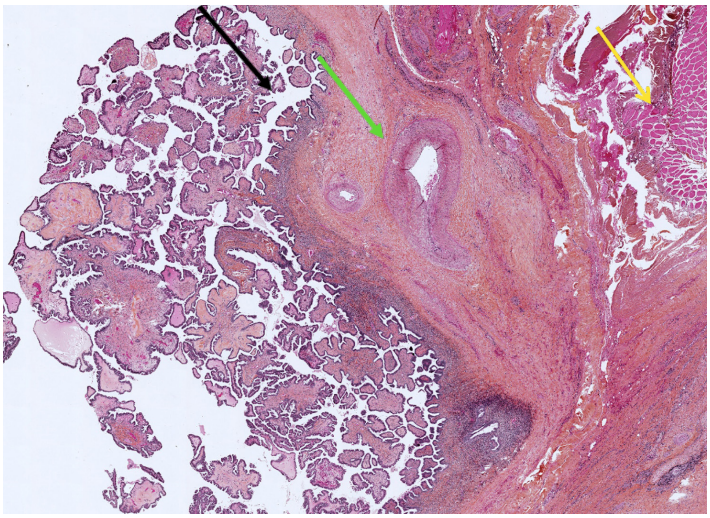


Figure 1: MRI images from October 2017: axial and sagittal sections showing retroperitoneal cystic lesion **(A),** para-aortic lymph node **(B).**



Microscopic view of the serous borderline ovarian tumor (arborescent, complex papillary architecture) (black arrow); in the middle (green arrow) the large vessels and on the right at the top (yellow arrow) a normal striated muscle.

Figure 2: Renal fascia showing two superficial non-invasive micro-foci of a papillary tumor: microscopic photo of a Hematoxein-Eosin-Safran (HES) stain enlarged 20 times.

The patient was subsequently referred to the Department of Gynecology-Obstetrics of Tenon Hospital where she had been treated for a right BOT in 2011. The BOT had been discovered incidentally during laparoscopy for infertility. Biopsies of exophytic ovarian vegetations revealed a serous BOT with micropapillary pattern. Serum tumor markers revealed elevated CA125 at 91 IU (N<35) with normal serum levels of CEA and CA 19-9. MRI showed an epithelial ovarian lesion measuring 5.7x5.5 cm with exophytic vegetations as well as a suspicion of peritoneal implants in the vesico-uterine fold and the Pouch of Douglas associated with an 18 mm retroperitoneal cyst behind the kidney on behalf of a benign renal lesion. On perfusion MRI sequences, the mass exhibited a type 2 curve (Figure 3). The Apparent Diffusion Coefficient was 2.1 (without restriction). The patient underwent a laparoscopic staging including a right salpingo-oophorectomy, infra-colonic omentectomy and peritoneal biopsies with peritoneal washing. Histopathology showed a stage IIIA serous BOT without microinvasion but with micropapillary features, non-invasive peritoneal implants and positive peritoneal cytology. Omentectomy and diaphragmatic biopsies were negative. Restaging surgery was recommended because of the peritoneal implants. In April 2011, a CT scan confirmed the presence of a benign retroperitoneal cyst and revealed an unexpected ongoing intra-uterine pregnancy. In accordance with the patient's wishes, the restaging surgery was delayed until after the first trimester of pregnancy. The surgery revealed no residual intra-abdominal implant but the retroperitoneal space was not explored. The peritoneal cytology was negative. The patient was lost to follow up after giving birth in December 2011. Examination of the patient was normal in February 2018 with negative PET-FDG scan, tumor markers and diagnostic hysteroscopy. After concertation at a multidisciplinary oncology meeting, radical surgery by laparoscopy was recommended including hysterectomy, left salpingo-oophorectomy, pelvic and para-aortic dissection. Histology revealed the presence of a 1.8 cm left ovarian serous BOT without microinvasion or micropapillary features. Para-aortic lymphadenectomy found a serous BOT in one of 28 lymph nodes but without invasive features. The peritoneal cytology was negative. The oncology committee recommended expectant management and no recurrence has been reported 12 months after the radical surgery.

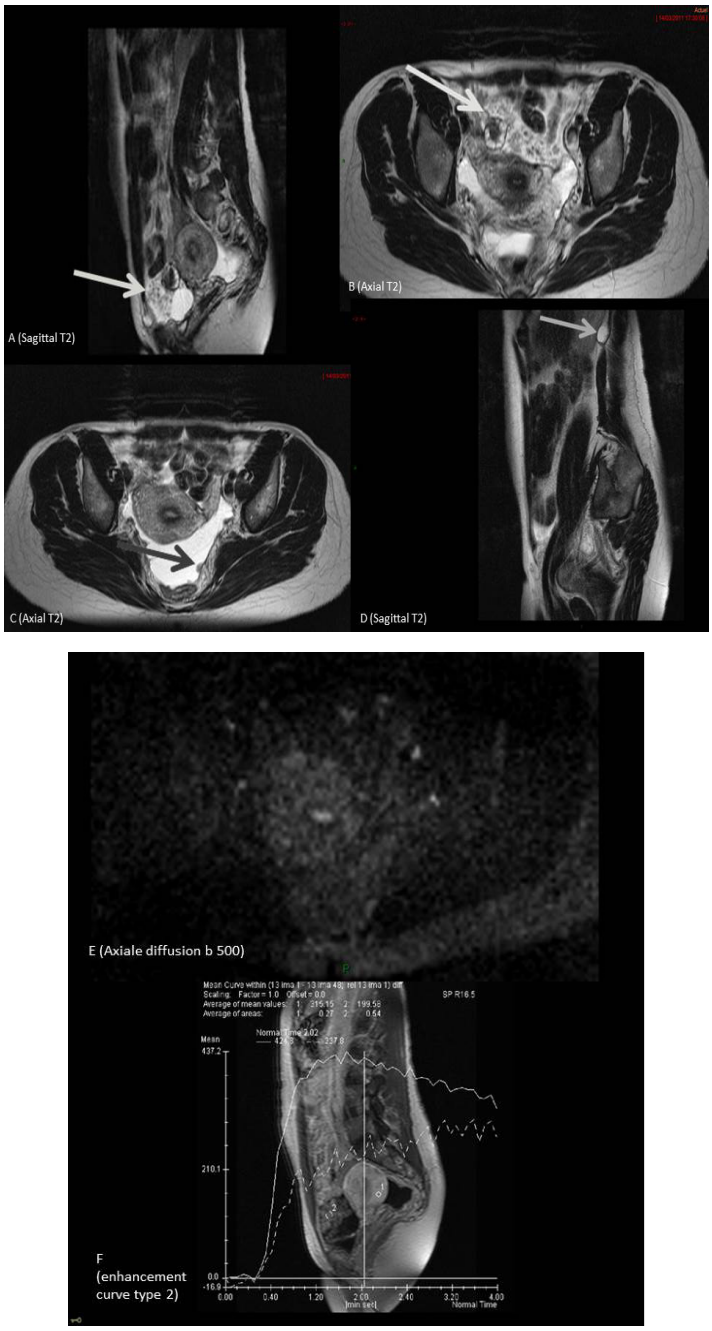


Figure 3: MRI images from February 2011: epithelial ovarian lesion with exophytic vegetations in sagittal and axial sections **(A,B),** peritoneal implants of the Pouch of Douglas **(C)** and an 18 mm retroperitoneal cyst behind the kidney representing a benign renal lesion **(D), E** (Axial diffusion b 500), **F** (perfusion curve type 2).

Discussion

Recurrence of a serous BOT in an extra-lymph node retroperitoneal location is a very rare condition. In the current case report, the main issue was to determine whether retroperitoneal space involvement was concomitant or not to the serous BOT. The absence of para-aortic lymph node on initial preoperative imaging was because of an independent retroperitoneal renal tumor exhibiting features of a benign tumor justifying expectant management. However, the subsequent development of the retroperitoneal tumor with a suspicious para-aortic lymph node was related to a malignant tumor of tubo-ovarian origin confirmed by histology. This location of serous BOT has only previously been reported in two articles, including five patients, underlying the rarity of this case. Rota, et al. [8] described three patients with serous BOT with microscopic nodal involvement and one retroperitoneal recurrence in a serous BOT. Shiraki, et al. [9] described one case of lymph node excision for a serous BOT (FIGO Stage III C) associated with extensive ovarian external papillary growth, peritoneal implants in the omentum and Pouch of Douglas, and involvement of multiple pelvic and paraaortic lymph nodes. In a series of 81 women undergoing surgical staging including retroperitoneal sampling for BOT, three patients (3.7%) with serous tumor had microscopic nodal involvement [8]. Among 236 patients with BOT, but without systematic lymph node sampling, only one retroperitoneal recurrence (0.4%) of a serous tumor was observed [8].

As mentioned above, the crucial issue in our patient was to determine whether the retroperitoneal tumor was a synchronous tumor or a secondary location after nodal involvement. For mucinous tumors several hypotheses have been suggested to explain the retroperitoneal location such as ectopic ovarian tissue, teratoma in which the mucinous epithelium prevails over all other components, intestinal duplication (also known as enterogenous genesis) [10-12], as well as invagination of the peritoneal mesothelial layer with subsequent development of a cyst [13,14]. The only features suggestive of a retroperitoneal location of serous BOT are the presence of ectopic ovarian tissue and the invagination of peritoneal implants. The concomitance of both a serous BOT and a retroperitoneal cystic lesion in our patient pleads in favor of ectopic ovarian tissue. Conversely, the micropapillary pattern of the serous BOT with peritoneal implants supports retroperitoneal diffusion by contiguity of an initial misdiagnosis of intraperitoneal disease. Indeed, some peritoneal surfaces such as the pre-renal fascia are difficult to fully explore by laparoscopy [15]. Moreover, this hypothesis is consistent with the presence of non-invasive micro-foci of a papillary tumor on the renal fascia. Finally, the last hypothesis is a retroperitoneal metastasis subsequent to an initially misdiagnosed para-aortic lymph node metastasis. Invasive peritoneal implants for serous BOT and residual disease after surgery are the two clearly identified factors to define a high-risk group of recurrence [1]. Other factors are controversial for increased risk of invasive recurrence; micropapillary patterns in serous BOT, intraepithelial carcinoma in mucinous BOT, stromal microinvasion, and conservative treatment based on cystectomy [1,6,16-20]. The presence of micropapillary pattern, observed in 10–15% of serous BOT, increases the likelihood of both invasive peritoneal implants and recurrence [21]. Moreover, although an initial unilateral salpingo-oophorectomy was performed, contralateral recurrence might also contribute to retroperitoneal recurrence.

In summary, our case report demonstrates that serous BOT can occur in an unusual distant location despite the low malignancy potential. This implies that comprehensive preoperative imaging should be systematically performed to distinguish synchronous from recurrent BOT. Furthermore, it raises the issue of restaging surgery for serous BOT with incomplete initial intra-peritoneal exploration especially when exhibiting micropapillary pattern.

Conclusion

In summary, this case demonstrates that serous BOT can arise in distant locations despite its low potential for malignancy. Imaging, requiring biopsies and restaging surgery, plays a crucial role in both detection and diagnosis. Due to the low recurrence rate and the possibility of delaying removal without negatively impacting overall survival, fertility sparing surgery should be discussed in young patients wishing to preserve their childbearing potential.

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