

A Rare Report of Concurrent Serous and Mucinous Cystadenomas in Bilateral Ovaries

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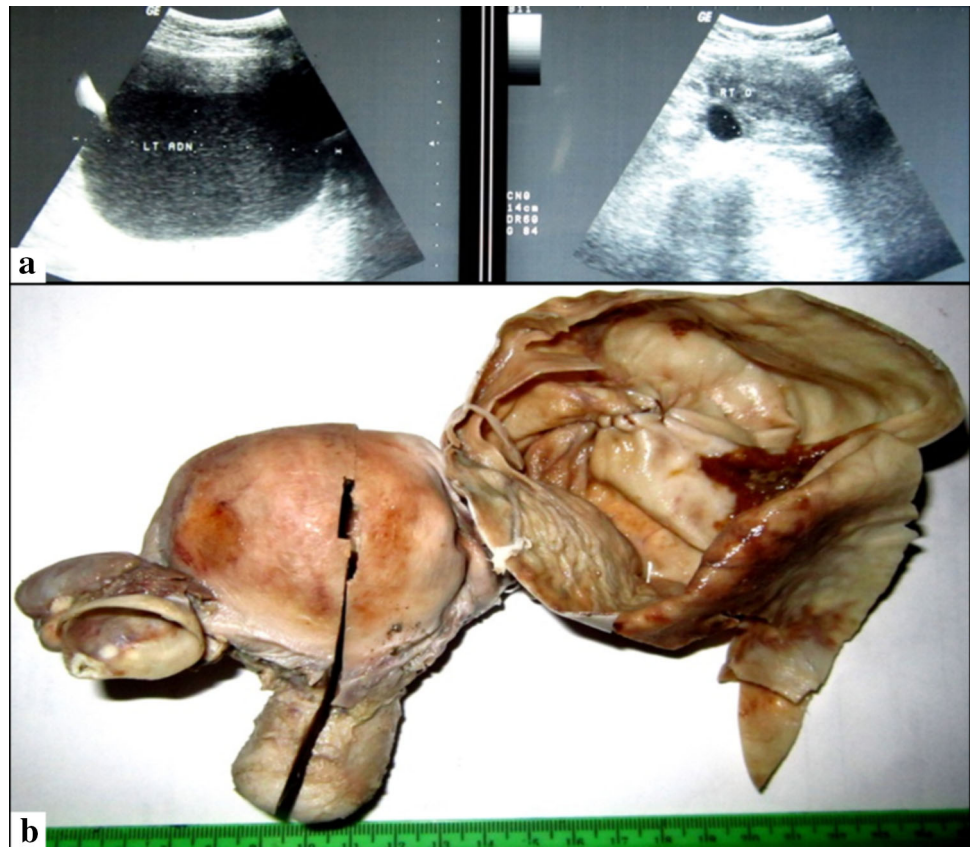
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Introduction

Epithelial tumors in the ovary primarily originate from its surface epithelium or any of its inclusion cysts. These tumors comprise about 60% of all ovarian neoplasms. The epithelium has the potential to differentiate into serous, mucinous, endometrioid, clear cell, or transitional epithelium, and thus any benign, borderline, or malignant tumor thereof. Serous and mucinous tumors are its commonest derivatives, which together reckon for approximately 30% of all ovarian neoplasms. Around 25–50% of the serous tumors are bilateral, most of which are borderline tumors (SBT). But simultaneous involvement of both ovaries with primary tumors of different histology is highly unusual [1].

Fig. 1 Sonographically, left ovarian cystadenoma with small right ovarian cyst (**a**). Macroscopically, thin-walled multiloculated cysts in left ovary with a simple cyst on right side (**b**)



In this latest presentation, we highlight a case of bilateral ovarian cystadenomas of serous and mucinous histotypes. To our deepest knowledge, this case is only the second of its kind to be described in the literature.

Case History

A 55-year-old postmenopausal woman presented with vague lower abdominal pain since past 7 months. Her abdomino-pelvic palpation and per-vaginal examination failed to detect any relevant pathology. Pelvic ultrasonography reported a normal endo-myometrial echotexture, with conjugal presence of a left ovarian cystadenoma and another simple right ovarian cyst (Fig. 1a). Serum CA-125 was estimated as 12 U/ml (normal: <35 U/ml). Subsequently, the patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy.

Grossly, both the ovaries were cystically enlarged—left > right. Their surfaces appeared smooth and glistening white. The left ovary measured around 10 × 6 × 5 cms. On resection, multiple well-delineated cysts were visualized, which expressed thin and clear transudate fluid. The right ovary measured approximately 3 × 2 × 2 cms. Its cut section revealed a uniloculated cyst with focal

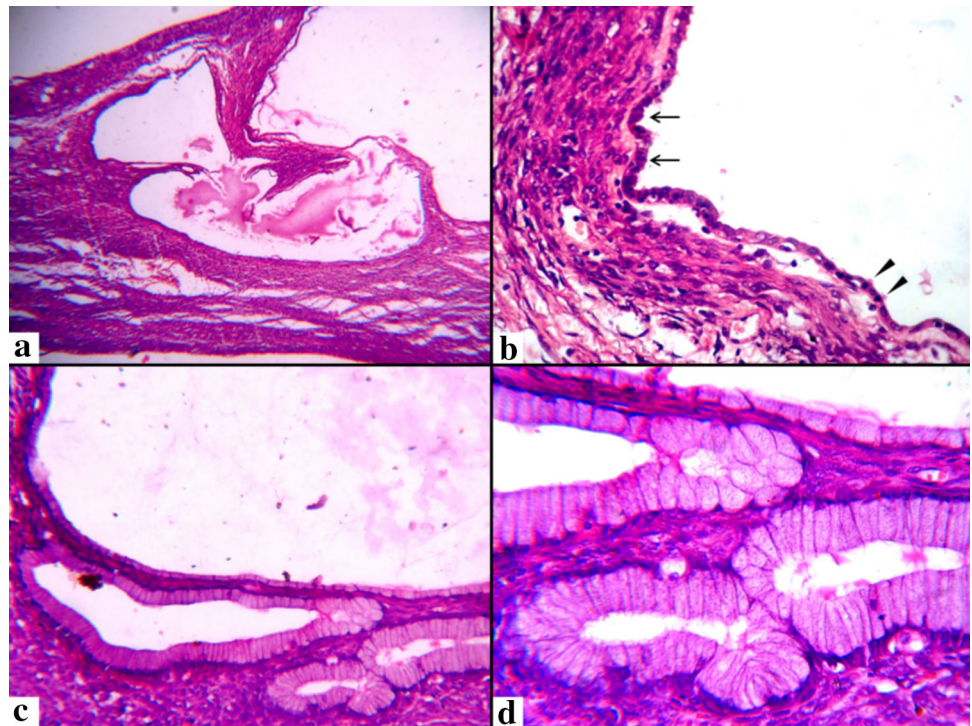
thickening of its inferolateral wall (Fig. 1b). The cyst was filled up with glutinous mucoid fluid.

On histopathology, the left ovarian cysts were lined by single-layer tubal-type epithelium. It exhibited the characteristic presence of secretory cells with elongated nuclei and ciliated cells with rounded nuclei. At places, the epithelium was also flattened. Straightaway, the diagnosis was serous cystadenoma. On the other hand, the right-sided cyst was lined by mucinous columnar epithelium with their nuclei pushed toward the base. The thick part of cyst wall bore multiple closely apposed small mucus glands, dispensed within cellular fibrotic stroma (Fig. 2). Therefore, it was diagnosed as mucinous cystadenoma. None of the cyst epithelia demonstrated nuclear stratification, atypia, or mitoses, which eventually potentiated the benign nature of both cysts. Postsurgically, the patient experienced an uneventful recovery and was absolutely symptomfree since 6-month follow-up.

Discussion

Primary bilateral ovarian involvement is most commonly seen with the serous neoplasms. For invasive micropapillary serous carcinoma (MPSC), bilaterality is observed to be highest, i.e., about 80–90%. This is followed by

Fig. 2 Microscopically, left ovarian serous cystadenoma (a H&E stain, $\times 40$), lined by an epithelium resembling the fallopian tube with ciliated (arrow-heads) and non-ciliated cells (arrows) (b H&E stain, $\times 400$); right ovarian mucinous cystadenoma (c H&E stain, $\times 100$), lined by mucin-secreting columnar epithelium and its wall containing several small mucinous glands (d H&E stain, $\times 400$)



noninvasive MPSC and SBTs. Bilateral involvement is present in two-third and one-third cases of these tumors, respectively. Other less frequent epithelial tumors to produce bilateral ovarian masses include endometrioid tumors, clear cell tumors, adenofibromas, Brenner tumors, and seromucinous tumors [1]. But a primary bilateral ovarian neoplasm exhibiting two different epithelial components in either site is extremely unusual. In 2014, Preeti et al. [2] reported a patient with synchronous enlargement of both ovaries with serous and clear cell carcinomas. Later Sule et al. [3] encountered a bilateral ovarian neoplasm with germ cell components of mature teratoma and mucinous cystadenoma. In this context, simultaneous development of serous and mucinous cystadenomas in bilateral ovaries, exactly similar to the discussed case, has been described just once before [4]. Therefore, this currently observed case stands only the second of its example.

In the identical report as the present one, Sethi et al. [4] encountered focal nuclear stratification, papillary excrescences and mild-to-moderate nuclear atypia in the otherwise benign serous cystadenoma. Diffuse presence of these features is actually indicative of SBTs, and an additional presence of destructive stromal invasion favors serous carcinoma. A quite indifferent criterion is applied for the mucinous tumors also [1]. However, the tumors from our discussed case exclusively exhibited the benign histomorphology of serous and mucinous neoplasms. Even after extensive serial-sectioning, none of the ovaries did manifest any dysplastic changes at all.

Among the patients suffering from gynecological neoplasms, approximately 1–2% females harbor the tumor simultaneously at two or more primary genital sites. Concurrent endometrial and ovarian malignancy is its commonest example. Any other combinations, including independent primary involvement of bilateral ovaries like the present case, are much infrequent. These coincidental tumors may express an entirely different histomorphology, or an identical one. This latter situation puzzles the diagnostic scenario when all the synchronous tumors are malignant. Such a presentation strongly favors metastasis from one to another location [1]. On the other aspect, concurrence of two different epithelial morphologies in unilateral ovarian neoplasm results from divergent differentiation of the neoplastic clone. But when it appears in either ovary, the histogenesis includes: (1) Two different primaries developing synchronously, or (2) Divergent differentiation of one malignancy after metastasizing to contralateral ovary [2]. However, in case the concurrent tumors are exclusive of benign morphology, as observed in the current case also, the former histogenetic pathway poses to be the most appropriate one.

In conclusion, bilateral concurrent benign ovarian tumors of different epithelial morphologies are extremely uncommon. Only a handful number of cases have been described in the literature. But still its incidence rate might have been little under-estimated due to the lack of thoroughness applied for routine histopathological examination of an apparently uninvolved ovary.

Compliance with Ethical Standards

Conflict of interest Krishnendu Mondal, Tarak Banik and Rupali Mandal declare that they have no conflict of interest.

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