

## CASE REPORT

# Coexistence of Benign Brenner Tumor with Mucinous Cystadenoma: A Case Report

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### ABSTRACT

Brenner tumor (BT) is a rare type of surface epithelial neoplasm found in the ovaries, accounting for just 2% of all ovarian tumors. They are typically diagnosed in post-menopausal women in their fifth to sixth decade of life. The occurrence of BT in young adults is uncommon. The coexistence of BTs with other surface epithelial tumors is unusual. The tumors are usually small, solid, unilateral and detected incidentally however, when they contain cystic components, they can appear larger. The proposed histogenesis is that they stem from urothelial metaplasia of the ovarian surface epithelium. There are only a very limited number of published case reports that discuss benign BT coexisting with a mucinous cystadenoma of the ovary in young adults. Therefore, we are presenting this case report, involving similar tumor in a 30-year-old unmarried woman.

**Keywords:** Brenner tumor, epithelial ovarian tumor, mixed epithelial tumor, mucinous cystadenoma, ovarian cyst.

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### INTRODUCTION

Ovarian tumors represent a significant health concern for women, leading to both mortality and morbidity. These tumors display a wide range of histological types and are categorized into 13 distinct groups by the World Health Organization.<sup>1</sup> Among these categories, epithelial ovarian tumor is the most common and are further classified into five subtypes due to diverse histogenetic origin: serous, mucinous, endometrioid, transitional, and clear cell.<sup>1</sup> Among these subtypes, BTs are relatively less common, accounting for approximately 2% of epithelial tumors.<sup>2</sup> BTs are frequently encountered as incidental findings.<sup>2</sup> These tumors are characterized by their fibro-epithelial nature, and feature a transitional cell appearance within the proliferating epithelial component, indicative of metaplasia.<sup>2</sup>

Mucinous neoplasms are the largest, but less frequent than serous tumors and accounting for 10-15% of ovarian tumors, and are categorized as benign, borderline, or malignant.<sup>1,3</sup> The majority (80%) of benign mucinous neoplasms fall into the categories of mucinous cystadenoma and mucinous adenofibroma.<sup>3</sup> When a tumor contains small areas of a different tumor type in addition to the main one, these minor foci are typically disregarded.<sup>3</sup> However, if a significant portion (more than 10%) of multiple tumor types coexist within the same ovarian tumor, it is

described as a mixed epithelial tumor.<sup>3</sup> Approximately 20% of BTs can present with other epithelial tumors and germ cell tumors such as mucinous cystadenoma, serous cystadenoma, benign cystic teratoma, or struma ovarii.<sup>4</sup> Roma AA and Masand RP reported up to 27% of BTs associated with mucinous tumors.<sup>5</sup>

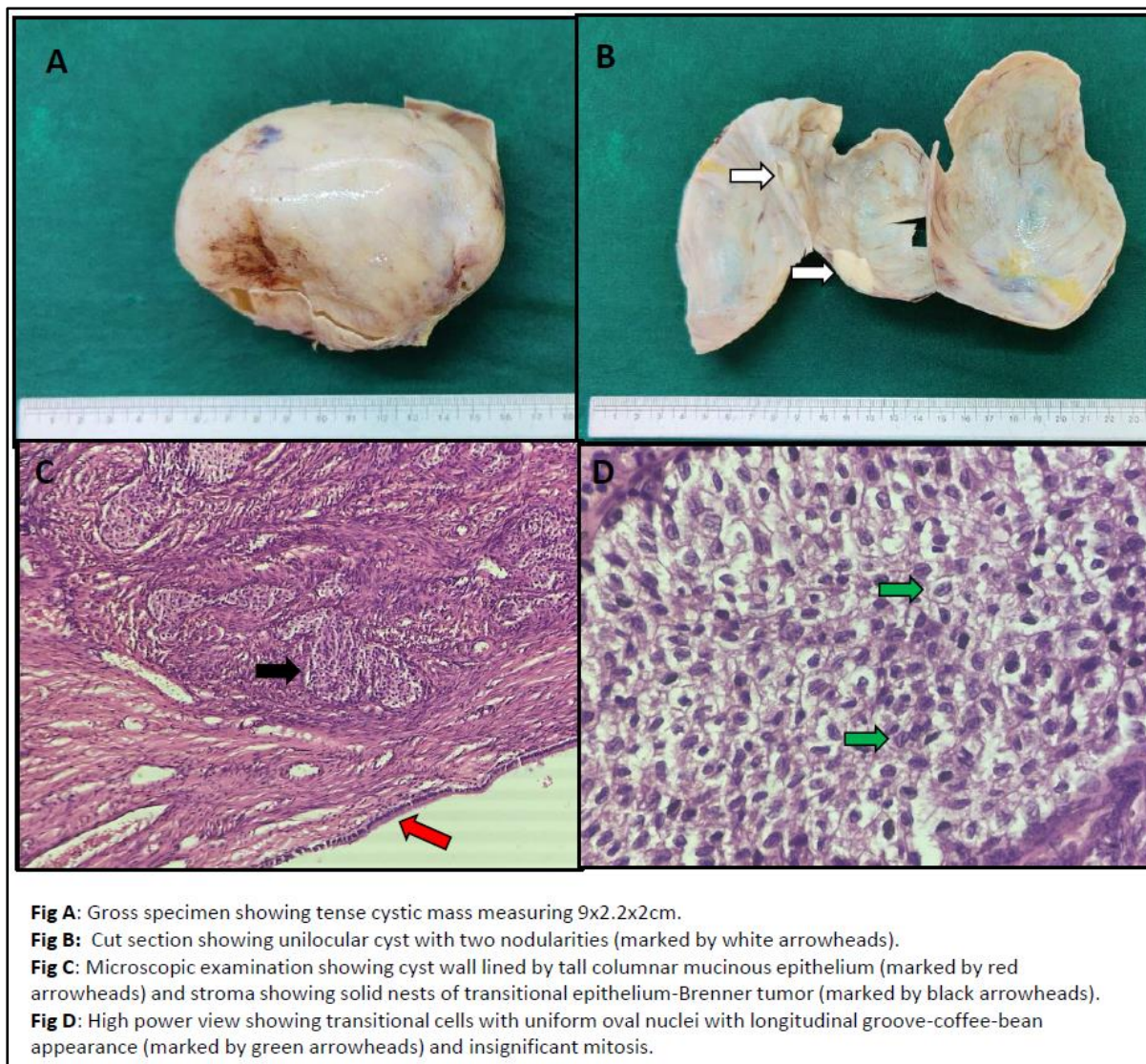
### CASE REPORT

A 30-year-old unmarried lady presented with a history of mass abdomen for 1 year. There was no history of any major disease or prior surgeries. Ultrasonography of the abdomen and pelvis revealed a large right-sided ovarian cyst. The patient underwent a right-sided oophorectomy and the specimen was for histopathological examination in 10% formalin.

The specimen received comprised of tense cystic mass measured 9x2.2x2cms and weighed 275gms. The external surface showed a tense capsule and congested blood vessels. The cut section showed it to be a unilocular, focally thick-walled cyst, measuring 9cm in diameter and filled with serous fluid. The inner lining was smooth in most areas and showed two focal nodularities, larger measuring 2.5x2cm and smaller measuring 2x1cm, respectively. Cut section through the nodular areas shows greyish-white solid areas. No papillary excrescences, areas of necrosis, or hemorrhage were seen.

Microscopic examination from multiple sections of the cystic mass showed the cyst wall to be lined by tall columnar epithelium with nuclei located towards the base. No necrosis or hemorrhage were seen. It contained intracellular pale-staining mucin and showed Periodic acid-Schiff stain (PAS) positivity and PAS-diastase (DPAS) resistance. Sections from the nodular areas showed nests of tumor cells containing

oval to polygonal epithelial cells that resembled urothelial cells in a background of abundant fibrous stroma. These cells exhibited a moderate amount of clear to eosinophilic cytoplasm and had round to oval bland vesicular nuclei, some of which had grooving nuclei. Hence histological features are consistent with mixed epithelial tumors with components of BT and mucinous cystadenoma.



## DISCUSSION

BTs are a rare subtype of epithelial surface tumors, constituting only about 5% of benign ovarian epithelial tumors.<sup>2</sup> They usually develop in adults during their fifth to seventh decade of life, in post-menopausal women.<sup>2</sup> This case, the patient was a 30-year-old young female.

They are generally asymptomatic, but occasionally symptoms such as abdominal pain, vaginal bleeding, or the presence of a palpable mass are observed in patients with BT.<sup>6</sup> In this case, the patient presented with an abdominal mass.

BT (95%) are unilateral and are typically discovered incidentally during pathological

examination.<sup>2,7</sup> This patient also demonstrated this unilateral characteristic. These tumors are generally small, measuring less than 2cm in size, and have a solid, firm, greyish-white appearance.<sup>2</sup> However, when associated with mucinous or serous cystadenoma, they can present as larger masses, as was the case with our patient, whose tumor measured 9 cm in diameter. Brenner tumors are often seen as small mural nodule, which was also observed in this case.<sup>6</sup>

Mucinous ovarian tumors comprise a smaller proportion of epithelial ovarian tumors than serous tumors.<sup>1,3</sup> The majority (80%) of the mucinous ovarian

lesions are benign.<sup>1</sup>Majority(95%) present as unilateral and with an average size of 10cm.<sup>1</sup>

Histologically, BT can be benign, borderline, or malignant.<sup>2</sup>They are characterized by the presence of sheets of nests, that resemble those found in the urinary bladder, forming both solid and cystic epithelial nests surrounded by a dense fibroblastic stroma.<sup>2,8</sup>The cells within these nests exhibit oval nuclei with distinct nucleoli and frequent nuclear grooving.<sup>2,8</sup> The coexistence of epithelial and stromal components is considered a defining feature of the BT.<sup>2</sup> The cysts associated with these tumors are lined by metaplastic columnar epithelium and often contain eosinophilic secretions within the lumen, suggesting features of mucinous cystadenoma.<sup>2</sup>The same characteristics were observed in this patient's case.

A hypothesis suggests that BTs originate from cells located at the tubo-peritoneal junction, which go through a process of transitional cell metaplasia before they invaginate the paratubal or ovarian surface, forming structures known as Walthard nests.<sup>3,7</sup> It is postulated that these Walthard nests might eventually seed the ovary and develop into BT.<sup>3,4</sup> Hence the coexistence of mucinous cystadenomas and BT in the ovary lends support to the theory that these two types of tumor may share a common origin, possibly arising from either the ovarian surface epithelium or remnants of the embryonic mesonephric system.<sup>9</sup>

In comparing our case with other reported cases, the mixed epithelial tumors of BT with mucinous cystadenoma were observed in the elderly, postmenopausal age group.<sup>10-12</sup> In contrast, this case involved a young reproductive female. Furthermore, the disease course in the reviewed case reports was of shorter duration (less than 6 months), while this patient's presentation was longer duration (more than a year).<sup>10-12</sup>Clinical presentation across various cases commonly included abdominal distension and discomfort, a pattern consistent with this patient's symptoms. Additionally, an enlarged size of the ovary was seen in all cases, aligning with the presentation of this report.<sup>10-12</sup>In all case reports, total abdominal hysterectomy with bilateral salpingo-oophorectomy was the surgical intervention.<sup>10-12</sup> In this case, only oophorectomy was done.

These distinctions emphasize the unique aspects of this case, particularly regarding the presentation in young reproductive age, longer duration of symptoms, and the surgical approach of oophorectomy chosen.

## CONCLUSION

This case is reported to create awareness among pathologists and gynecologists regarding the coexistence of ovarian mucinous cystadenoma and Brenner tumors in young women. It's important to note that both of these tumor types are benign, and the standard treatment involves cystectomy, which is curative. However, it is essential to be vigilant when dealing with mucinous cystadenoma cases because

they have been known to recur after cystectomy, primarily due to the potential rupture and spillage of their content.

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