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Bilateral Ovarian Gonadoblastoma Associated With Foci Of Dysgerminoma : A Case Report And Literature Review

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Abstract

Introduction

Ovarian gonadoblastoma is a rare germ cell tumor commonly associated with dysgenetic gonads and the presence of Y chromosome material. Although generally considered histologically benign, it carries a substantial risk of malignant transformation, most frequently into dysgerminoma.

Case Presentation

We report the case of a 35-year-old woman presenting with bilateral ovarian masses initially suspected to be cystic lesions. Histopathological examination revealed bilateral ovarian gonadoblastoma associated with foci of dysgerminoma. This case is remarkable because of the patient's adult age, bilateral ovarian involvement, and the absence of apparent phenotypic abnormalities.

Conclusion

This case highlights the importance of systematic histopathological examination of ovarian masses, even when they appear clinically benign. It also emphasizes the need for multidisciplinary management and long-term follow-up because of the risk of malignant transformation and recurrence.

Keywords : bilateral ovarian tumor; dysgerminoma; germ cell tumor; histopathology ; ovarian gonadoblastoma.

Introduction

Gonadoblastoma is a rare germ cell tumor of the gonads characterized by a mixed proliferation of germ cells and gonadal stromal cells. First described by Scully, it preferentially develops in dysgenetic gonads and is strongly associated with the presence of Y chromosome material [1]. Although histologically considered benign, gonadoblastoma has a high potential for malignant transformation, mainly into dysgerminoma, occurring in more than 50% of reported cases [2].

Dysgerminoma represents the most common malignant tumor associated with gonadoblastoma and corresponds to the ovarian counterpart of testicular seminoma [3]. This association confers particular clinical significance to gonadoblastoma, requiring appropriate management and close follow-up.

Most reported cases involve young patients presenting with disorders of sexual differentiation or gonadal dysgenesis [4]. However, several cases occurring in phenotypically normal women have also been described, suggesting variability in the underlying pathophysiological mechanisms [5].

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In this context, we report a case of bilateral ovarian gonadoblastoma associated with foci of dysgerminoma in a 35-year-old woman and discuss its clinicopathological features in light of the current literature.

Case presentation

A 35-year-old woman was admitted for pelvic symptoms leading to the discovery of bilateral ovarian masses initially suspected to correspond to symptomatic ovarian cysts.

Gross examination revealed three non-oriented whitish nodular masses. The largest lesion measured 14.5 × 10 × 7 cm, was well encapsulated, and had an elastic consistency. On cut section, it showed a heterogeneous gelatinous appearance with brownish, greenish, and whitish areas. The two additional masses were partially encapsulated and measured 10 × 6 × 2 cm and 5.5 × 4.5 × 2 cm, respectively. They displayed similar morphological features, including cystic cavities.

Histopathological examination demonstrated a tumoral proliferation composed of solid nests and trabeculae of immature cells. Tumor cells exhibited rounded nuclei with variable cytoplasm and were arranged within a fibrous stroma that was frequently thickened. Lymphocytic infiltrates, spindle-shaped stromal cells, and multiple calcification foci were also identified (Figure 1). Residual ovarian stroma was observed at the periphery.

These morphological findings were consistent with gonadoblastoma. In addition, some areas exhibited proliferation of atypical germ cells compatible with dysgerminoma, confirming the coexistence of both entities.

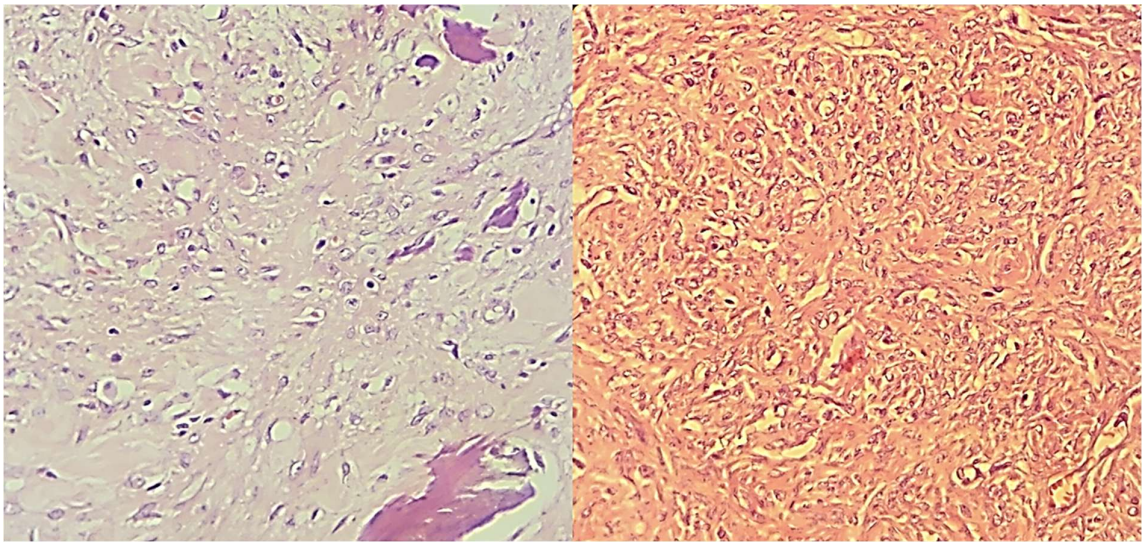

Figure 1 : Ovarian resection specimen showing histological features of gonadoblastoma associated with dysgerminoma. Hematoxylin and eosin stain x 400. Source : Department of Pathology and Cytology, University Hospital Andrainjato, Fianarantsoa, Madagascar

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Discussion

Gonadoblastoma is a rare tumor whose pathogenesis is closely associated with the presence of abnormal germ cells in the setting of gonadal dysgenesis. According to Scully, it is a mixed tumor partially reproducing the organization of embryonic gonads [1].

The presence of Y chromosome material is considered a major factor in tumorigenesis. The TSPY gene located on the Y chromosome plays a significant role in germ cell proliferation and neoplastic transformation [6]. However, the absence of clinically apparent abnormalities in some patients, as observed in the present case, suggests that additional mechanisms, including environmental or epigenetic factors, may also contribute to tumor development [5].

Dysgerminoma is the malignant tumor most frequently associated with gonadoblastoma and results from the malignant transformation of tumoral germ cells [3]. Several studies have reported this transformation in more than half of the cases [2].

Histologically, gonadoblastoma is characterized by three major components: germ cells, stromal cells, and calcifications. Calcifications represent an important diagnostic feature reflecting degenerative intratumoral changes [7]. The lymphocytic infiltrate observed in our case has also been frequently described in the literature.

The particularity of the present observation lies in the bilateral involvement of the ovaries. All three surgical specimens demonstrated similar morphological features, consisting of gonadoblastoma associated with foci of dysgerminoma, indicating bilateral and multifocal ovarian disease. This presentation is consistent with the report by Arafa et al., who described bilateral ovarian gonadoblastoma with predominant dysgerminomatous proliferation in the right ovary in a 17-year-old patient with Swyer syndrome [8]. Although bilateral lesions have been reported, they remain relatively uncommon and may suggest a diffuse pathological process related to abnormal gonadal development [4].

Unlike the patient reported by Arafa et al., who presented with 46,XY gonadal dysgenesis, our patient was a 35-year-old phenotypically normal woman without reported developmental abnormalities. Another comparable case was reported by Yilmaz et al. involving a 20-year-old woman with bilateral ovarian gonadoblastoma associated with dysgerminoma [9]. Their observation closely resembles the present case because of both the bilateral ovarian involvement and the coexistence of gonadoblastoma and dysgerminoma.

However, the age of our patient is relatively advanced compared with most cases reported in the literature, which generally involve adolescents or young adults investigated for amenorrhea, pubertal developmental abnormalities, or pelvic masses. In the present case, the tumor was initially interpreted as a benign cystic ovarian lesion, demonstrating that gonadoblastoma may clinically mimic benign ovarian pathology.

Raafey et al. also described a rare case of bilateral gonadoblastoma associated with dysgerminoma in a phenotypically normal female with a 46,XX karyotype [10].

Similarly, Yin et al. reported ovarian gonadoblastoma associated with dysgerminoma in an 11-year-old girl with a 46,XX karyotype who presented with acute abdominal pain secondary to tumor rupture [11]. Although this pediatric case differs from the present observation in terms of age and clinical presentation, it confirms that gonadoblastoma associated with dysgerminoma may occur even in the absence of detectable Y chromosome material. Their report also underlines the importance of endocrine and genetic investigations, as the patient was ultimately diagnosed with 17α-hydroxylase/17,20-lyase deficiency [11]. In our case, cytogenetic or molecular investigations could also be considered to identify a possible occult chromosomal or endocrine abnormality.

Compared with previously published reports, the present case demonstrates three notable features: the adult age of the patient, the bilateral ovarian involvement, and the absence of apparent phenotypic abnormalities. Most similar cases described in the literature concern younger patients with gonadal dysgenesis or identifiable chromosomal abnormalities.

This observation demonstrates that gonadoblastoma associated with dysgerminoma should remain a differential diagnosis in cases of heterogeneous bilateral ovarian masses, even in adult women without obvious disorders of sexual

development. It also highlights the importance of multidisciplinary management involving pathology, gynecologic oncology, endocrinology, and, whenever possible, cytogenetic evaluation.

Failure to establish an early diagnosis may adversely affect patient outcomes, emphasizing the importance of systematic histopathological examination of ovarian masses, including lesions initially considered benign.

Conclusion

Ovarian gonadoblastoma is a rare tumor with significant malignant potential. The present observation highlights a bilateral form associated with dysgerminoma in an adult woman without apparent phenotypic abnormalities, an uncommon clinical presentation.

This case emphasizes the crucial role of histopathological analysis in the diagnosis of ovarian masses and underlines the importance of appropriate multidisciplinary management and long-term follow-up.

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