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A Case of Papillary Thyroid Carcinoma Arising from Struma Ovarii and Extending into the Bladder

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Abstract

Ovarian goiter is a form of single tissue teratoma of the ovary, accounting for 2% - 3% of mature ovarian teratomas. Malignant transformation may occur in rare cases. Papillary thyroid-type carcinoma represents the most common type of malignant struma ovarii, followed by follicular carcinoma. Malignant struma ovarii is commonly seen in women in the fifth decade. The diagnosis is often made post-operatively after histological examination. Histology also helps assess tumor aggressiveness (mitoses, necrosis, poorly differentiated subtype, etc.). Given the rarity of these lesions, no therapeutic consensus or prognostic value had yet been formally established. We report herein, the case of a 76-year-old woman with a cystic tumor of the right ovary and a nodular lesion of the bladder. The clinical symptomatology is nonspecific, associating abdomino-pelvic pain and a right latero-uterine mass on abdominal palpation. After total hysterectomy with bilateral adnexectomy, the diagnosis of papillary carcinoma arising from struma ovarii and extending into the bladder was made. Through this observation, we suggest to discuss the anatomoclinical particularities of this rare pathological entity.

Keywords

Struma Ovarii, Ovary, Papillary Carcinoma, Thyroid, Bladder Nodule

1. Introduction

Ovarian goiter or struma ovarii is a monodermic teratoma of the ovary, mainly (>50% of the tumor) or exclusively composed of thyroid tissue [1]. This rare

clinical entity represents 2% - 3% of mature ovarian teratomas [1] [2]. Malignant transformation can be found less frequently and metastases remain exceptional. Since the earliest descriptions of struma ovarii by Von Kalden in 1895 and Gottschalk in 1899, there is a large discrepancy in the reported frequency of the malignant form of these tumors due to their rare nature and the lack of standard diagnostic criteria [3]. The most common type of malignancy observed is papillary carcinoma followed by follicular carcinoma [1] [2] [3]. Clinical manifestations of malignant struma ovarii are usually nonspecific, characterized by the presence of an abdomino-pelvic mass or an abdominal distension and signs of hyperthyroidism can be encountered in rare cases. These malignant tumors are often only diagnosed post-operatively [4] [5].

Herein, we report a case of papillary thyroid carcinoma arising from a struma ovarii and involving the bladder.

Our purpose was to highlight through this case the anatomo-clinical particularities and the histologic criteria for the diagnosis of this rare and incidentally discovered lesion.

2. Observation

A 76-year-old women, postmenopausal and multiparous with no significant past medical history, presented for abdomino-pelvic pain, evolving for several months. On physical examination, the patient was in good general condition. The presence of a right latero-uterine mass was found on vaginal examination combined with abdominal palpation. The remainder of the physical examination was unremarkable. An abdomino-pelvic ultrasound was performed, showing a multilocular cystic ovarian mass, measuring 12 cm of long axis with heterogeneous components (**Figure 1**). There was no evidence of peritoneal effusion. The dome of the bladder presented a hyperechoic formation measuring 3 cm in long axis in contact with the right ovary. A cystoscopy was then performed, showing a normal-looking bladder mucosa, with no specific lesion. In addition, biological assessment, including serum levels of tumor markers (CA125: 15 U/ml), was normal. After a multidisciplinary team meeting, a total hysterectomy with bilateral adnexectomy and resection of the bladder nodule were performed.

Surgical specimens were sent for pathological examination. On macroscopic examination (**Figure 2**), the right ovary measured 11 × 8 × 6 cm with a smooth grayish-brown external surface of renitent consistency. On opening, there are solid greenish gelatinous areas, sometimes calcified, and multilocular cystic areas, 1 to 5 cm in diameter, with yellowish liquid content. The bladder nodule, also grayish-brown, measured 3 × 1.5 × 1 cm and cut sections showed a single cystic cavity, containing pilo-sebaceous elements. On histological examination, the ovarian mass is basically composed of thyroid follicles, varying in size, containing colloid (**Figure 3(A)**). Foci of calcification were often identified. Thyreocytes demonstrated within a focus of 15 mm, papillary-type nuclear atypia, such as overlapping ground glass nuclei, irregular nuclear contour or nuclear grooves (**Figure 3(B)**). The wall of the cystic cavity of the bladder nodule was lined with

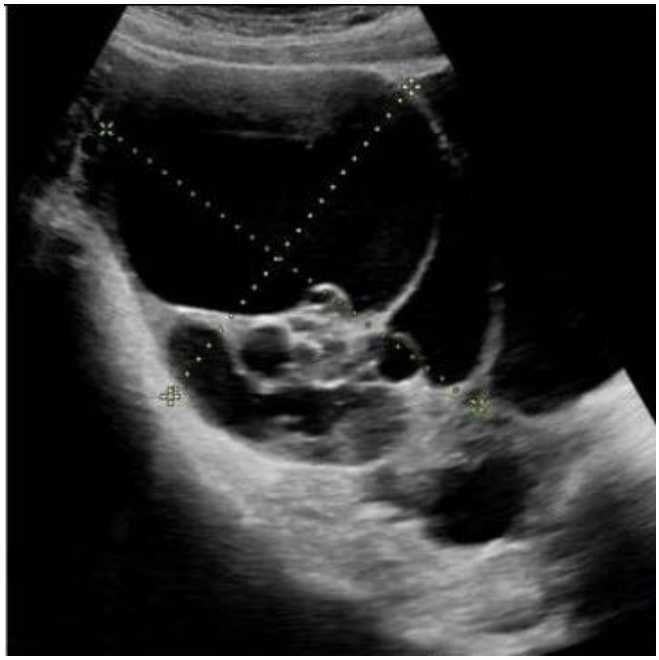

Figure 1. Ovarian mass with cystic and solid components on ultrasound. Source: Radiology Department, CHU Andrainjato Fianarantsoa, Madagascar.

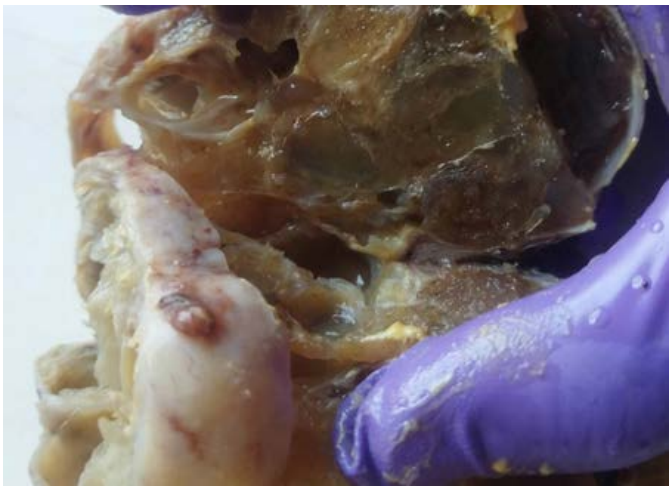

Figure 2. Right ovarian mass with multiloculated cyst and solid gelatinous materials.

squamous stratified epithelium. Adnexal structures like hair follicles and pilo-sebaceous units were seen as well as thyroid follicles. Follicular cells also demonstrated equivocal features of papillary carcinoma as described previously. The diagnosis of thyroid papillary carcinoma arising from struma ovarii with bladder extension was then made. An abdomino-pelvic CT scan was performed for an extension assessment, showing no metastatic location (**Figure 4**). Besides, thyroid hormonal status was also evaluated and was in normal range. No additional treatment was performed. Clinical evolution was favorable. The patient was in good general condition and was free of recurrence at 6 grayish-brown month follow-up.

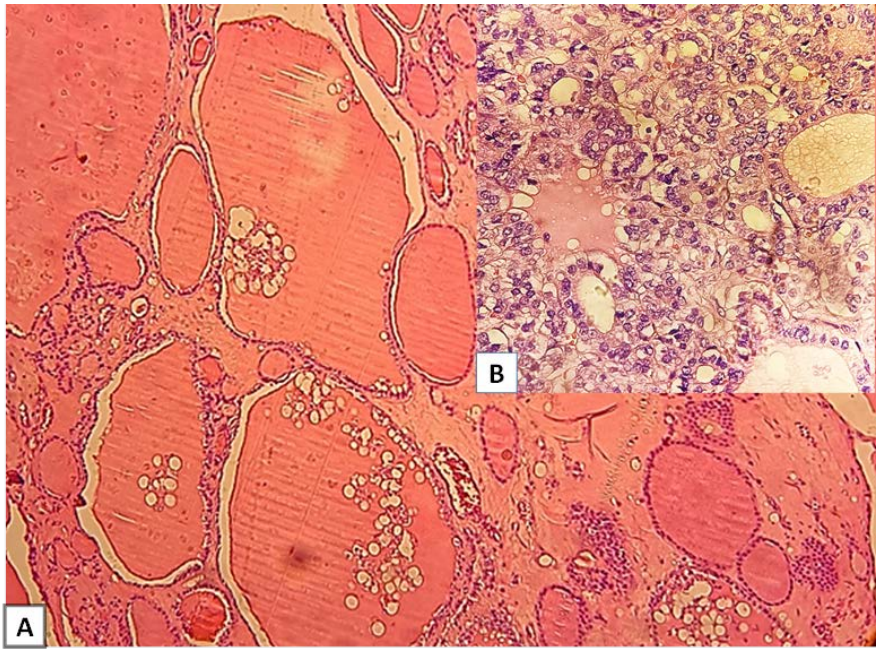

Figure 3. Malignant struma ovarii. A, Thyroid follicles varying in size, containing colloid (Hematein-eosin stain × 200). B, focus of papillary thyroid carcinoma (Hematein-eosin stain × 400).

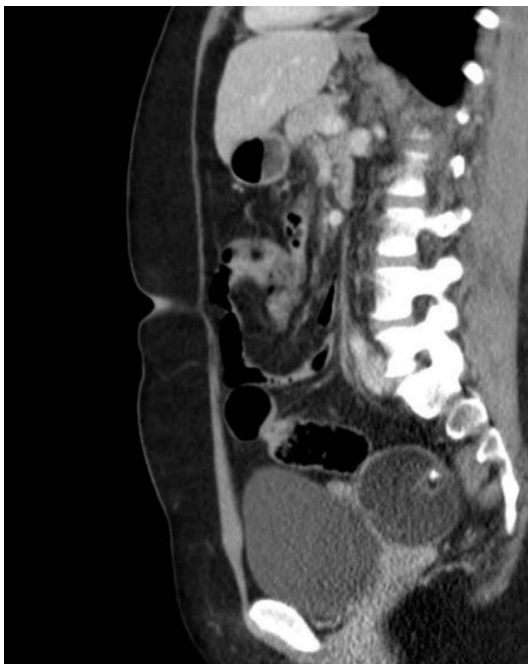

Figure 4. Abdomino-pelvic CT scan showing no metastatic location. Source: Radiology Department, CHU Andrainjato Fianarantsoa, Madagascar.

3. Discussion

Ovarian goiter is a monodermic teratoma of the ovary, mainly or exclusively composed of thyroid tissue that may develop in rare cases malignant transformation [1] [2]. Malignant struma ovarii is mostly encountered in women in the

fifth decade and is usually associated with a papillary thyroid carcinoma. The exact etiopathogenesis remains unclear [1] [2] [3] [4]. In our case, the patient was 76 years old and had no particular medical history. Since these malignant forms very rarely metastasize, the clinical symptomatology is non-specific and generally associates pelvic pain, abdominal mass, pseudo-Meigs syndrome (ascites, hydrothorax) and more rarely hyperthyroidism [6]. Complications may occur such as infection, torsion, compression, rupture, or even fistula formation into neighboring organs, accompanied by urinary or digestive signs or peritonitis [5] [6]. Imaging studies could not determine the presence or absence of malignant component. Ultrasound shows a heterogeneous lobulated mass with multilocular cyst and solid components [2] [6]. MRI gives more specific images, showing multi-partitioned mass with variable signals, often with a marked hyposignal on T1 and T2, related to the presence of colloid [6] [7]. For our patient, the clinical presentation was essentially characterized by abdomino-pelvic pain and a right latero-uterine mass on abdominal palpation. The ultrasound revealed the ovarian origin of the mass and the presence of a nodular formation of the bladder dome in contact with the ovarian mass. The diagnosis of struma ovarii is based on histopathological examination and only pathological results could reveal the presence of malignant transformation [7] [8]. Histological diagnosis of struma ovarii is usually easy. It corresponds to the presence of thyroid follicles, containing colloid, resembling that of normal thyroid gland [9]. For the diagnosis of malignant struma ovarii, the histologic criteria are still controversial. Some authors have suggested that nuclear atypia alone is not a sufficient diagnostic criteria. They required the presence of capsular invasion and metastasis or peritoneal implant [1] [6] [10]. Besides, many other authors have argued that the diagnosis of malignancy can be made on the basis of cellularity, cellular atypia, mitotic activity and vascular invasion can be a helpful feature but not obligatory [11]. There are also disagreements regarding the architecture of these lesions. The older literature categorized predominantly follicular patterned tumors as follicular carcinomas. However, more recent papers have shown that tumors with a follicular architecture but with psammoma bodies, infiltrative growth pattern and papillary carcinoma cytologic features behave as papillary carcinoma (regional node metastases) and not as follicular carcinoma (bone and hematogenous metastases) [12]. This could suggest that the diagnosis of malignant struma ovarii should be rendered using the histopathological criteria for primary thyroid carcinoma. Furthermore, another form of challenging behaviours of these tumors has been also described. This consists of benign struma ovarii without malignant features that developed recurrences or metastases several years after the initial diagnosis. Recurrences can show benign appearing thyroid tissue, features of papillary or follicular carcinoma [10] [11] [12]. Metastatic spread usually follows that of ovarian cancer. Most common sites are contralateral ovary and more distant sites such as bone, liver, brain and lungs [6] [11] [12]. Bladder involvement as described in our case is less common. For our patient, histological features of papillary thyroid carcinoma with follicular patterns

have been found. Thyrocytes exhibited papillary type nuclear atypia including nuclear grooves, ground glass nuclei, irregular nuclear contour. The presence of similar malignant components within the bladder nodule led to the diagnosis of bladder extension. However, the presence of associated benign features such as hair follicles and pilo-sebaceous units within the bladder nodule still remains difficult to explain although capsular effraction was not observed. Histopathological analysis could also help assessing tumor aggressiveness, such as: a poorly differentiated architecture (trabecular, solid, insular), high mitotic activity, pattern of tumor necrosis or presence of vascular emboli, carcinoma component > 4 cm, an ovarian capsule rupture, a neuro-endocrine component, ovarian serosal implants, presence of thyroid tissue or neuro-endocrine component on peritoneal samples [6] [9]. Immunohistochemistry can sometimes be helpful to confirm the presence of thyroid tissue (TG, TTF1) or the existence of a neuro-endocrine component (chromogranin A, synaptophysin) [6]. In our case, the presence exclusively of thyroid follicles in the ovarian mass and the features of papillary thyroid carcinoma within the ovary and the bladder nodule led to the diagnosis of malignant struma ovarii with bladder extension. The papillary carcinoma did not present any feature of aggressiveness and was limited to 15 mm within the ovary. Due to the low prevalence of malignant struma ovarii, no therapeutic consensus exists. Management strategies should then be defined on the basis of a multidisciplinary consultation. For young patients who wish to preserve their fertility, in the absence of contralateral involvement, the treatment can generally correspond to a cystectomy or a unilateral salpingo-oophorectomy with peritoneal cytology for evaluating tumor spread. For elderly or menopausal patients as in our case, the treatment is not standard but may consist of a bilateral oophorectomy with bilateral adnexectomy and total hysterectomy [6] [13]. Additional treatment is sometimes necessary and depends on tumor extension [13]. Assessment of tumor extension could help to rule out ovarian metastasis from a primary thyroid tumor, bilateral ovarian locations or multifocal metastatic location. In the absence of aggressiveness characteristics or if the tumour is completely excised, a simple pelvic ultrasound as well as a thyroid ultrasound may be sufficient. Conversely, the extension assessment will be completed by an abdomino-pelvic CT scan. Thus, radioactive iodine therapy I-131 can be offered as a complementary treatment after thyroidectomy for aggressive or metastatic tumors [6] [14]. For our patient, the extension assessment did not show any metastatic localization and no adjuvant treatment had been performed. Malignant struma ovarii often have a good prognosis with a low risk of recurrence. For metastatic forms, their prognostic value has not yet been established due to the rarity of these lesions [6]. In our case, the evolution was favorable, with no signs of recurrence or residual tumor at 6-month follow-up.

4. Conclusion

Malignant struma ovarii constitutes a rare pathological entity, which is often diagnosed postoperatively during histopathological examination. There are still

some difficulties in the diagnosis of these tumors due to the lack of standard histologic criteria. In our case, the diagnosis of malignancy was based on the foci of marked papillary-type nuclear atypia in the ovary and the bladder nodule. In all cases, these tumors have a better overall prognosis with unpredictable behavior and possible recurrences that do not correlate with the morphology.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

Patient Consent

The patient has given written informed consent to publish the present manuscript.

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