

# Papillary Carcinoma on Stroma Ovarii: A Case Report from Madagascar and Review of the Literature

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## Abstract

A 64-year-old woman with no particular history presented with chronic pelvic pain since November 2021. The ultrasound performed showed multiple right ovarian cystic ranging from 0.4 to 4 cm on the long axis. She underwent a right salpingo-oophorectomy in May 2022. The anatomical pathology result is in favor of a papillary carcinoma developed on stroma ovarii. No adjuvant treatment was necessary since the tumor was well limited, without capsular rupture, stage IA. The thyroid test was normal. The patient is currently being monitored. There is no sign of recurrence 20 months after surgery.

## Keywords

Papillary Carcinoma, Surgery, Struma Ovarii

## 1. Introduction

Mature ovarian teratoma is composed mainly or exclusively of thyroid tissue. It represents less than 3% of mature teratomas, 5% to 10% of ovarian goiters, and 0.01% of ovarian tumors [1]. Ovarian struma, first reported by Bottlin in 1888 and later by Pick in 1902, is a rare monodermal variant of ovarian teratoma [2]. A malignant tumor on the ovarian struma is a rare tumor with one case reported in Madagascar [3]. The management strategy is based on whether ovarian surgery is associated or not with radioactive iodine [4]. Patients with malignant ovarian struma had an excellent disease-specific survival rate [5]. The authors report here a case of papillary thyroid carcinoma occurring on an ovarian struma of the

right ovary in a 64-year-old woman.

## 2. Case Report

A 64-year-old woman, with no particular history, presented with chronic pelvic pain since November 2021. The pelvic ultrasound performed in April 2022 showed multiple cystic right ovarian from 0.5 to 4 centimeters in long axis (**Figure 1**). In May 2022, she underwent a right salpingo-oophorectomy. The pathological examination was in favor of a papillary thyroid carcinoma developed on stroma ovarii (**Figure 2**). The immunohistochemical examination was not carried out due to the patient's lack of financial means. The thyroid test was normal. The Cancer Antigen 125 (CA-125) level was normal at 18.9 U/ml. The thyroid ultrasound performed showed three hypodense, homogeneous nodules belonging to the benign series. The thoraco-abdominopelvic scan carried out in June 2022 did not show any residual or tumor recurrence. After discussion in the Multidisciplinary Consultation Meeting, clinical monitoring every 3 months and thyroglobulin dosage every 6 months were proposed. However, treatment with radioactive iodine would be initiated in the event of a recurrence. In December 2023, there are no signs of a recurrence.

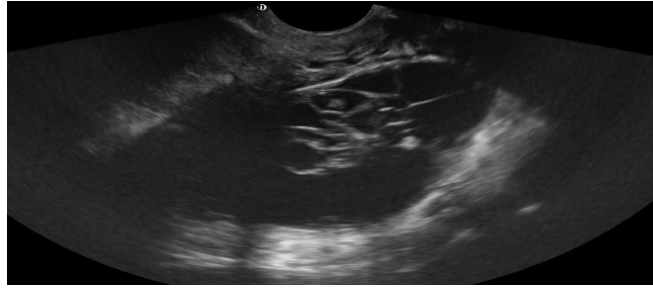
## 3. Discussion

Papillary carcinoma on the ovarian struma preferentially affects premenopausal women with an average age of around 42.5 years. Subjects aged over 60 years represent only 13% of patients [6]. The age of our patient was 67 years old.

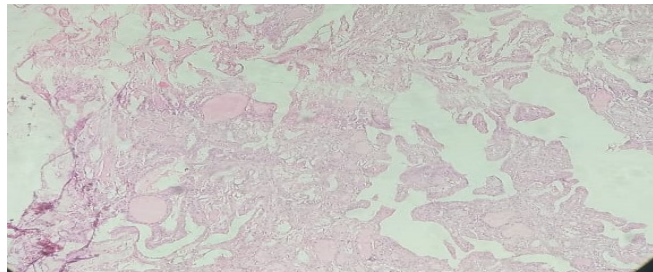
The clinical symptoms are nonspecific and similar to those of other ovarian neoplasms [4]. Depending on the size of the lesion, we may find an abdominal mass and pelvic pain. Our patient presented with chronic pelvic pain as her main symptoms. In rare cases or in advanced forms, pleural effusion and ascites are present.

Most cases occur on one side only. The frequency of bilateral disease was 5% [5]. Symptoms of hyperthyroidism are found in 5% - 8% of cases and result in a decrease in Thyroid-Stimulating Hormone level (TSH) and an increase in T3 and T4 sometimes associated with an increase in thyroglobulin [7]. The elevation of the tumor marker CA-125 is not associated with the malignancy of ovarian goiter and has little clinical value in the ovarian struma [8]. Imaging examinations cannot determine the presence or absence of a malignant component. Pelvic ultrasound reveals images in favor of a suspicious complex ovarian tumor: a large, multiloculated mass, with a predominantly cystic or mixed appearance [9].

No standard treatment has been established in the published literature. Total abdominal hysterectomy, bilateral adnexectomy and omentectomy are considered optimal [4]. Some reports have revealed that unilateral oophorectomy in the absence of capsular invasion is performed as in our case [10]. Our patient had undergone an oophorectomy, stage IA.



**Figure 1.** Multiple cystic right ovarian from 0.5 to 4 centimeters in long axis on ultrasound.



**Figure 2.** Cells with ground glass nuclei with hematoxylin and eosin staining.

Typical macroscopic examination shows a tumor with mixed solid and cystic components with mucosal or gelatinous content associated with mature teratoma components in the majority of cases. Histological diagnosis is based on the presence of ground glass nuclei, nuclear pseudo-inclusions and furrows seen on hematoxylin and eosin staining [3].

Papillary carcinoma is the most common histological type of stromal ovarii malignancy, accounting for approximately 50% of malignant struma ovarii cases [11]. Immunohistochemical stains for thyroid transcription factor-1 (TTF-1) and thyroglobulin can help confirm the diagnosis [3]. For our patient, the immunohistochemical examination could not be carried out due to lack of financial resources.

An adjuvant treatment modality with radioactive iodine may be offered in cases of residual or recurrent disease [9]. Performing a total thyroidectomy followed by radioactive iodine treatment is not recommended in the literature for malignant ovarian struma confined to the ovary [12].

Currently, there is no consensus on surveillance modalities [12]. The evaluation of thyroid assessments and imaging examinations are the main pillars of the follow-up strategy in carcinoma on struma ovarii. For our patient, clinical and imaging monitoring every 3 months and measure serum thyroglobulin every 6 months were proposed. However, treatment with radioactive iodine would be started in the event of a recurrence.

The prognosis for malignant struma ovarii is generally good if there is no metastasis. Long-term monitoring of patients with malignant struma ovarii is recommended. In Sijian Li's study, he recommends a follow-up of at least 20

years because the median duration of recurrence in their study was 14 years [13]. At 20 months of treatment, there is no sign of recurrence for our patient.

#### 4. Conclusion

Our observation highlights the importance of sending all the evidence to pathology even if the lesions appear clinically benign in order to know the prognosis and the most appropriate treatment.

#### Conflicts of Interest

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

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