

Ovarian Oxyphilic Sertoli Cell Tumor: Case Report And Review Of The Literature

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Citation

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Abstract

Background: the ovarian oxyphilic Sertoli cell tumor is a rare neoplasm (only three cases were reported in literature). Sometimes the rarity itself of a lesion may rise some problem in the diagnosis, especially if it is sent to several consultants.

Case: an unusual case of ovarian neoplasm came to our attention, and we sent some slides of the case to two renowned referees of different Departments of Pathology. Pathologist 1 made a diagnosis of endometrioid adenocarcinoma, while Pathologist 2 made the diagnosis of oxyphilic Sertoli cell tumor. He sends the same slides to Pathologist 1, who confirmed his diagnosis.

Conclusion: the two different diagnosis set different managements of the lesion for the clinician, but overall they set the pathologist who requested the consultation in a difficult position. In fact, when a pathologist sends to two experienced consultants a case, he really thinks to solve definitely the case itself. Unfortunately, sometimes it not happens.

INTRODUCTION

The oxyphilic Sertoli cell tumor is an extremely rare neoplastic pathology of the ovary. It was described for the first time from Ferry et al (1). In this paper they reported three young women (aged respectively 19, 21 and 30 years), two with the Peutz-Jeghers syndrome, which had unilateral ovarian tumors composed of Sertoli cells with abundant with eosinophilic cytoplasm ("oxyphilic"). The authors emphasized the need to include the Sertoli cell tumor in the differential diagnosis of oxyphilic ovarian neoplasms, particularly if there is a tubular pattern resembling the Sertoli structures. In the following years there wasn't any other report published in the literature about this tumor. In 1996 we observed a case of ovarian neoplasia simulating the oxyphilic Sertoli cell tumor. Since the lesion is extremely unusual, we decided to send in consultation to two expertise pathologists. When a "non specialist" pathologist has the diagnostic suspicion of an extremely rare lesion, it is a good way of thinking to send the histological slides and paraffin blocks to the pathologist which first described the lesion itself. Moreover, to obtain a conclusive confirmation it could be wise to send the case to another consultant with as expert in that field of pathology. The sending to two different

pathologists should obviously be limited to very selected cases. In almost all cases the answers of the consultants give us a precise, definite, conclusive diagnosis of the lesion, but sometimes as in the present case the answer could create a "hard" condition for the clinician and the pathologist who requested the consultation, condition that may take to an enigmatic, difficult management of the lesion itself.

PATIENT AND METHODS

A woman, aged 50 years, came to the attention of the gynecologist in 1997 for the incidental finding of a right ovarian mass during an abdomen ultrasonographic exam. After a laparotomy the surgeon performed an hysterectomy with bilateral salpingectomy. The right ovary weighted 210 gr, its main diameter was 16 cm with outer smooth, pinky surface. On the cut the mass was firm, yellowish, cystic centrally: the major thickness of the wall of the lesion was of 4 cm, and the inner surface appeared rough with yellowish, papillomatous proliferations, the major of the diameter of 0.8 cm. The left ovary appeared normal, and the uterus on the cut surface showed some leiomyomas of the anterior wall of the corpus and an atrophic endometrium. The microscopic examination of the ovarian mass showed a component of elongated tubules that grew in lobules separated by a typical

fibrous stroma, sometimes hyalinized. The tubules were lined by cuboidal cells, closely packed, with small nuclei and scanty cytoplasm, accompanied by a great component constituted by large cell with abundant, oxyphilic cytoplasm (figure 1-4). Our preliminary diagnosis was of “ovarian Sertoli cell tumor, oxyphilic cell variant”: we excluded the diagnosis of endometrioid adenocarcinoma (in our opinion the histological picture didn't fit with that diagnosis), and decided to send the histological slides to some consultants because of the rarity of the lesion. We choose a pathologist (Pathologist 1) of the Department of Pathology, Massachusetts General Hospital in Boston, and another pathologist (Pathologist 2) of the Department of Pathology and Laboratory Medicine (Anatomical Pathology), Vancouver Hospital, University of British Columbia, because of their high experience in the gynecopathologic field.

Figure 1

Figure 1: Ovarian Sertoli cell tumor (hematoxylin and eosin, 100x)

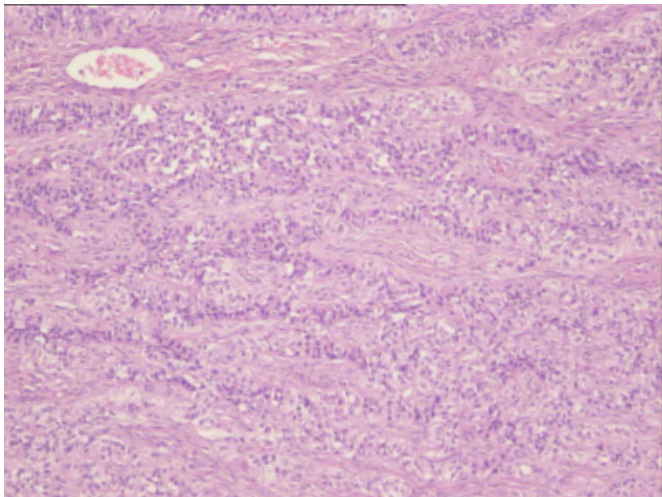


Figure 2

Figure 2: Ovarian Sertoli cell tumor (hematoxylin and eosin, 200x)

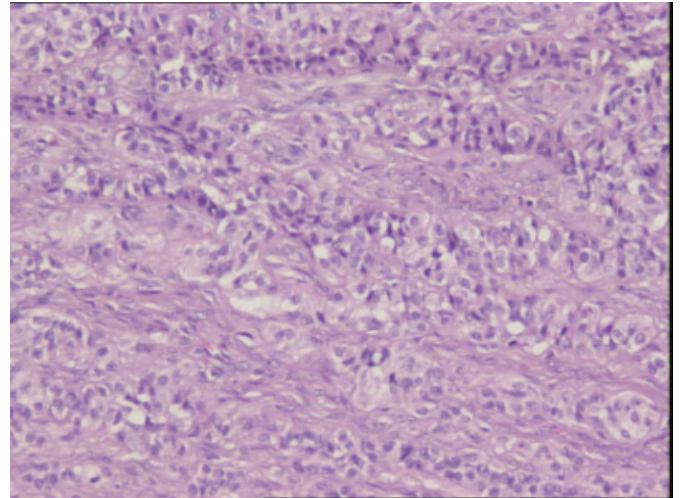


Figure 3

Figure 3: Ovarian Sertoli cell tumor: oxyphilic cells (hematoxylin and eosin, 200x)

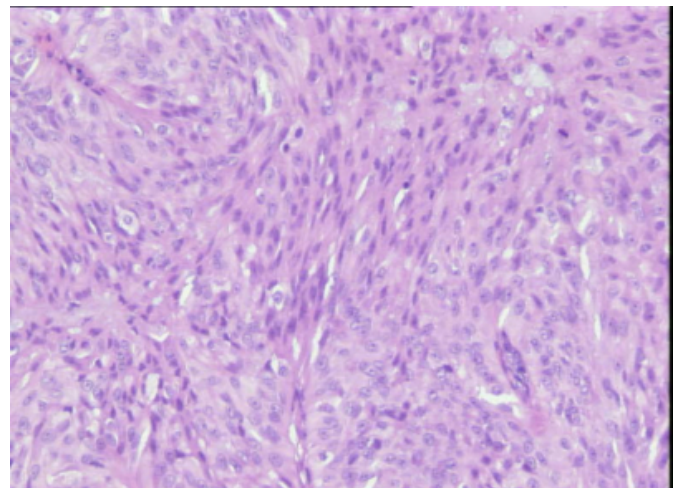
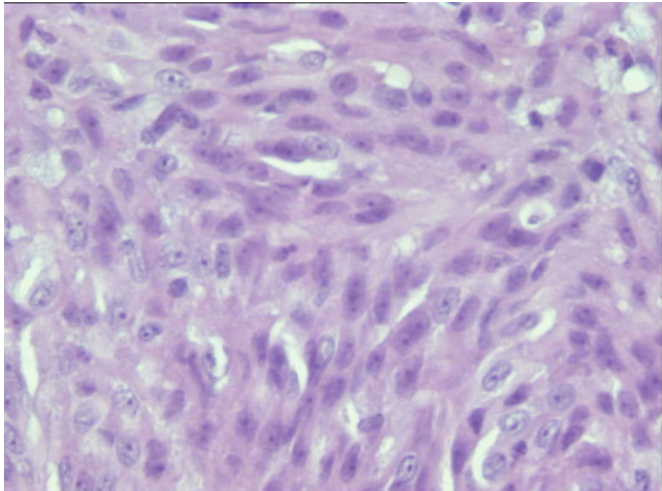


Figure 4

Figure 4: Ovarian Sertoli cell tumor: oxyphilic cells (hematoxylin and eosin, 400x)



At the present time after 8 years the patient is still alive, she never had any recurrence and is free of disease.

RESULTS

The answer of Pathologist 1 was the following: "This ovarian tumor in my opinion is a grade 2/3 endometrioid adenocarcinoma. There is abortive squamous differentiation and in many areas the sex cord-like formation one sees in many of these tumors." (bold characters by the writer). Pathologist 2 wrote: "I have reviewed the slides on the above case and performed immunohistochemical stains. It is indeed a challenging case. The tumor cells are characterized by abundant oxyphilic cytoplasm and appear to be growing in a variety of patterns including solid sheets and in other areas with striking sex-cord-like patterns reminiscent of that seen in a Sertoli cell tumor. My initial differential diagnosis was between an endometrioid carcinoma with sex-cord-like patterns and the rare oxyphilic Sertoli cell tumor which has been described in association with the Peutz-Jeghers syndrome. The immunohistochemical stains showing strong positivity for vimentin and keratin but negativity for epithelial membrane antigen supported a diagnosis of sex-cord tumor. I also referred some of the slides to Pathologist 1 and he agrees that that is the appropriate diagnosis in this case. The tumor in your case, therefore, should be considered to have a low malignant potential". (bold character by the writer).

DISCUSSION

A pathologist working in an institute to which comes any kind of lesions always has a limited experience in the field of extremely rare tumors. To avoid the misinterpretation of

rare pathologies calling them "usual" it is dutiful for the pathologist to send the histological slide to the first who described the lesion in literature and to another expertise in the same field of interest. With this procedure it is always to "solve" the enigmatic case, performing a right diagnosis for the clinician and the patient himself. But sometimes there shall be disagreement between the two consultants that diagnosed the lesion: in this time it arouse the problem, which one of the diagnoses shall the pathologist who requested the consultation choose.

In the present case our lesion was sent to two pathologists with great reputation in the field of gynecopathology, which often contribute each other between themselves. The diagnosis of ovarian oxyphilic Sertoli cell tumor was confirmed both Pathologist 1 than Pathologist 2, who sent the same slides to Pathologist 1. Still Pathologist 1 made a complete different diagnosis (ovarian endometrioid adenocarcinoma, grade 2-3) on the same material that we sent him: the histological slides sent to Pathologist 1 and 2 were identical; they were deep sections of the same histological paraffin block. The major problem that arises at this time is: because it is the requesting pathologist who has to make the final diagnosis, and just because it's him who has to talk to the clinician and be responsible of the diagnosis, which one of the two diagnoses shall believe? It is a very important problem for the clinician too, because depending on the diagnosis we give him he has to modify the therapeutic approach to the disease and the information to give to the patient himself, not only for the therapy but also for the prognosis.

The follow up of our case demonstrates that after 9 years the patient is still alive and free of disease, so it seems that the diagnosis of Pathologist 2, diagnosis that we choose, would be "correct" (although Ferry and coll. reported in their paper a recurrence in one of the three patients after 15 years from the surgical intervention). So the present case is the fourth case reported in literature of ovarian oxyphilic Sertoli cell tumor. It is also true that if a pathologist would make the "wrong choose" between two different diagnoses there will be a lot of consequences. Consequences would be not only by medical side (different therapy, different prognosis), but also by legal side (the patient could denounce the pathologist himself, the requesting pathologist, for inaccurate and wrong diagnosis). Our present case demonstrates so how for the expert pathologists too histopathology could sometimes be a real, surprising enigma.

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