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CLEAR CELL CARCINOMA AND ENDOMETRIOID CARCINOMA ARISING IN A ENDOMETRIOTIC CYST OF THE OVARY

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Synopsis A case of two tumors arising in an endometriotic cyst of the ovary is described. These two tumors are macroscopically polypous and are histologically a clear cell carcinoma and an endometrioid carcinoma, respectively. This is a rare neoplasm and its existence supports the general opinion that clear cell carcinoma and endometrioid carcinoma of the ovary are both müllerian in origin.

Key words: Clear cell carcinoma • Endometrioid carcinoma • Endometriosis • Malignant transformation of endometriosis • Ovary

Introduction

In 1925, Sampson[3] first reported some cases of malignant transformation arising in endometriosis and adopted the rigid criteria for this change. Corner et al.[2] and Scully et al.[7] reported that malignant change in ovarian endometriosis occurred in 6 of 889 cases (0.6%) and in 8 of 950 cases (0.8%) of endometriosis, respectively. Brooks et al.[1] reported malignant transformation arising in extragonadal endometriosis. They mentioned that various histological types of malignancy could arise in endometriosis.

Recently, the authors encountered a rare case, in which clear cell carcinoma and endometrioid carcinoma took place coincidently in a endometriotic cyst. The case is to be described in this report.

Case Report

The patient is a 36-year-old, single, gravida 2, para 0, woman. On October 8, 1984, she was admitted to the Hospital of Kumamoto University with sudden onset of lower abdominal pain. Physical examination: She was a well-developed, well-nourished woman, who was in moderately abdominal distress. Examination of the heart, lung, liver, spleen and kidney was almost normal findings. Slight deep tenderness on the lower abdomen. Pelvic examination and echography suggested that she had uterine myoma and bilateral ovarian tumors. Laboratory data on admission: Hematocrit 38%, Urinalysis: negative, Serology: negative, Serum CEA, hCG and AFP: negative, Chest X-ray: normal, EKG: normal. She underwent immediately a exploratory laparotomy. Examination of pelvic organs revealed that the uetrus was enlarged (new born child head size) and the both ovaries were enlarged (fist size) and adhered to the cul-de-sac. There were the blueberry-like lesions on the pelvic peritoneum. These findings were typical of endometriosis. She underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy.

Pathologic Observations

The macroscopic findings: The uterus weighed about 900g. Some myoma nodes were observed in the myometrium. The endometium was smooth, no apparant neoplastic growth was observed. The both ovaries were cystic, and weighed each about 200g. The both cyst were filled with a chocolate-like fluid. These cyst wall were thin, and these inside surfaces were generally smooth except for two localized solid tumors in the left ovary (Fig. 1). These tumors were gray-yellow, firm and polypous.

The microscopic findings: The myometrial nodes were leiomyoma. There was no malignancy in the endometrium. The cyst wall of both ovaries consisted of single layer of surface epithelium and the connective tissue, in which, hemosiderin laden macrophages were observed. There were two localized solid tumors in the left ovarian cyst. One of them was thumb head size (Fig. 2). The tumor cell nest showed solid or tubular proliferation, and divided by thin fibrous and hyalinous septa. The

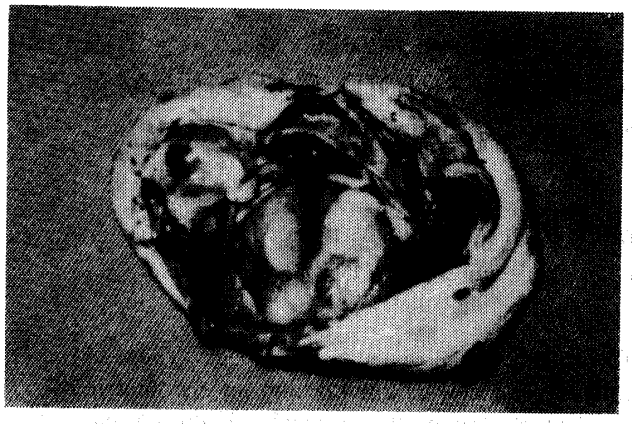

Fig. 1. Note two localized solid tumor in the left ovarian cyst. These tumor protrudes into lumen of cyst.

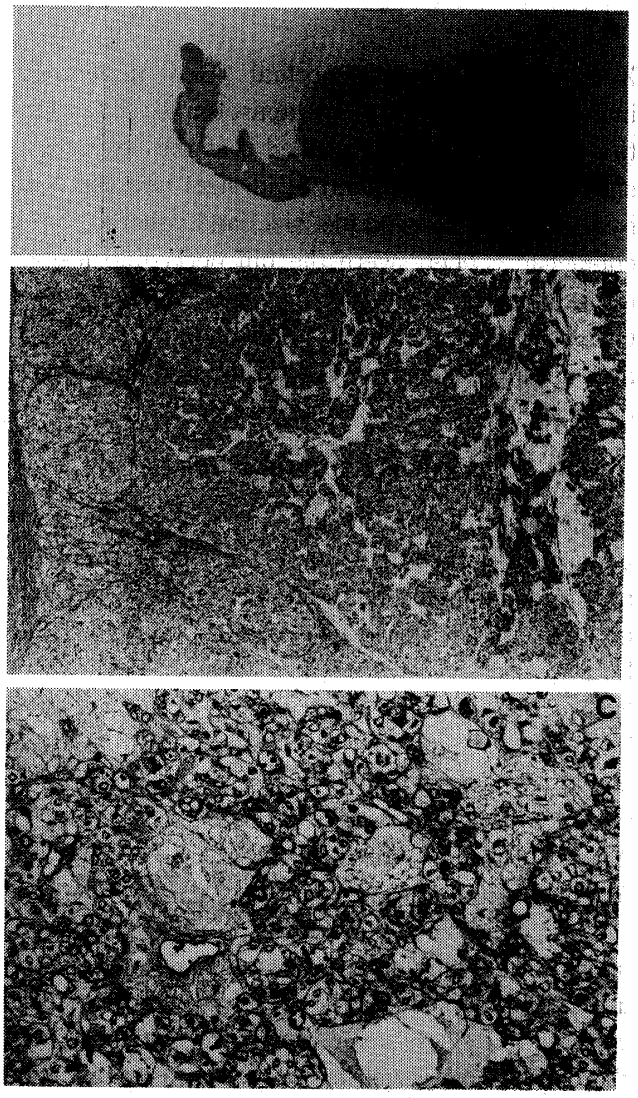

Fig. 2. A: 3×2×2cm sessile polypous tumor (2×). B: Note solid cancer cell nest with thin fibrous and hyalinous septa (20×). C: The tumor cells are polyhedral and had clear cytoplasm (200×).

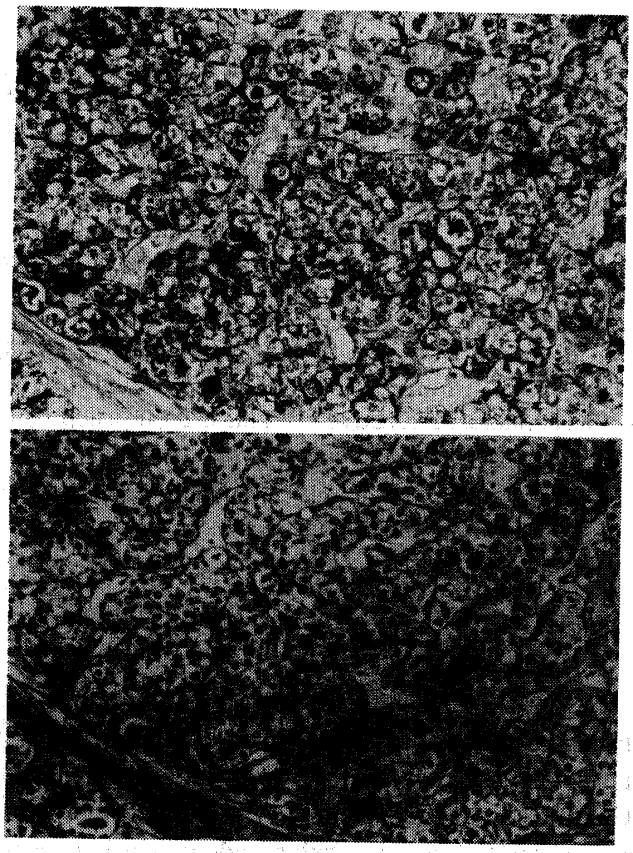

Fig. 3. A: The tumor cells are positively stained by PAS (200×). B: Negatively stained by diastase treated PAS (200×).

tumor cells had clear cytoplasm and large nuclei with prominent nucleoli and coarse chromatin net work. The tumor cells and secreting materials were postively stained with periodic acid Shiff (PAS). Then, the tumor cells were negatively stained with diastase digested PAS (Fig. 3). By these findings, this tumor was diagnosed clear cell carcinoma. Another tumor was little finger tip size and polypous (Fig. 4). The invasion into surrounding structure was unclear. The structure of this tumor was generally tubular and in part solid. These findings resembled that of endometrial polypoid neoplasm. This tumor cells showed loss of polarity and had irregular hyperchomatic nuclei. These finding suggested that this tumor was endometrioid carcinoma or endometrioid carcinoma of low malignant potential (LMP).

Discussion

As shown in Table 1[2] and 2[1], various histological types of malignancy can arise in ovarian endometriosis[8]. But, the majority of clinically

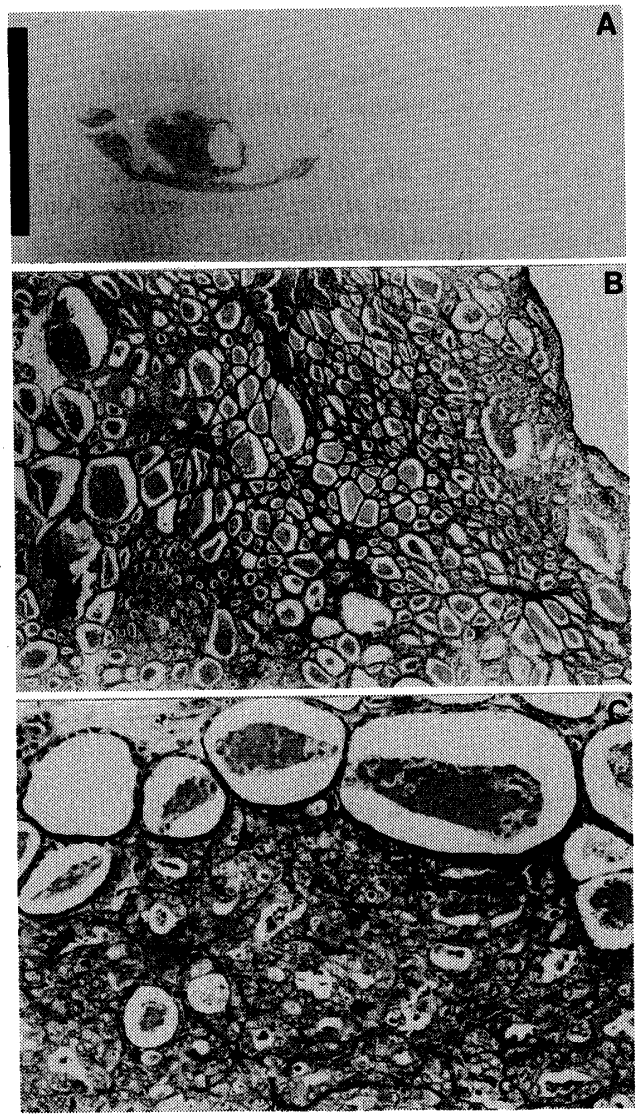

Fig. 4. A: 1×1×0.5cm pedunclar polypous tumor (2×). B: Tubular structure is prominent and is similar to the endometrial polyp (200×).
C: Note tubular and solid structure. The tumor cell nuclei are enlarged and had prominent nucleoi (200×).

discovered ovarian cancers are already in stage III or IV. It is difficult to decide whether malignant change in endometriosis can occur or not. Sampson[3] first advocated the rigid criteria: (1) Endometriosis and carcinoma co-exist in the same ovary. (2) The carcinoma is in the lesion of the endometriosis and not metastatic. (3) A transitional lesion is observed between the carcinoma focus and the endometriosis lesion. According to Scully et al.[7], the clinicopathological findings are as follows: (1) The tumor mostly results from an unilateral chocolate like cyst. (2) The tumor is generally projecting into the cyst cavity, but rarely out from the serosal side. (3) The tumor is frequently complicated by endometriosis in the pelvis and occasionally by endometrial carcinoma. (4) Most of the tumors are histologically classified as differentiated adenocarcinoma or adenoacanthoma.

Table 1. Ovarian carcinoma arising in endometriosis
(Corner G.W., Hu C.Y. and Hertig A.T.)

Histological type	No. of cases
papillary carcinoma	3
pseudomucinous cystadenocarcinoma	1
adenoacanthoma	1
serous cystadenocarcinoma	1

Table 2. Malignancy arising in extragonadal endometriosis (Brooks J.J. and Wheeler J.E.)

	Histological type	No. of cases
carcinoma	adenocarcinoma	21
	adenoacanthoma	7
	endometrioid carcinoma	2
	clear cell carcinoma	1
	squamous cell carcinoma	1
sarcoma	endometrial stromal sarcoma	10
	carcinosarcoma	1
	stromal endometriosis or low grade stromal sarcoma	2

The present neoplasm is arising in an unilateral chocolate like cyst, and the two tumor are polypously projecting into the cystic cavity. Macroscopically, in the pelvis, blueberry spots are present. Microscopically, this tumor is well differentiated adenocarcinoma. These tumors can be thought to occur probably from ovarian endometriosis, according to the Scully's findings.

One tumor is diagnosed endometrioid carcinoma or endometrioid carcinoma of low malignant potential (LMP). The diagnosis of endometrioid carcinoma (LMP) must be done carefully, and is difficult. Scully[5] mentioned that endometrioid carcinoma (LMP) was polypoid and adenofibroma or adenoacanthofibroma. The present tumor may be adenoacanthoma in situ.

Another tumor is diagnosed clear cell carcinoma. This diagnosis is reasonable by findings of hematoxylin-eosin and PAS-Diastase treated PAS stains. Since the report of Schiller in 1939[4], clear

cell carcinoma of the ovary have been considered by many investigators to arise from mesonephric remnants. But, in 1967, Scully and Barlow[6] presented convinsing evidence of the association of the so-called mesonephric carcinoma with endometriosis and endometriomata, suggesting the müllerian nature of this epithelium. It is now generally agreed that clear cell carcinoma of the female genital tract originate from müllerian epithelium. Ovarian clear cell carcinomas are relatively rare, comprising about 5% of all ovarian carcinomas. Most of them are probably derived from the surface epithelium, however, a small proportion of them are thought to arise from the epithelium of endometriotic cyst of the ovary.

We encountered a case of clear cell carcinoma and endometrioid carcinoma arising in a endometriotic cyst of the ovary. This case supports the concept that clear cell carcinomas and endometrioid carcinomas are of müllerian origin.

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概要 今回，私達は，一個の子宮内膜症性卵巣嚢胞内に発生した二個の腫瘍，すなわち，淡明細胞癌と類内膜癌を伴つた症例を経験したので報告する．子宮内膜症内に発生する悪性腫瘍は，種々の組織型が報告されているが，この症例のように二個の腫瘍が独立して，一個の嚢胞内に共存している例は稀である．また，この症例は，淡明細胞癌と類内膜癌がともにミューラー管由来であるという概念を支持する症例として，特異であると考えられる．