

## CASE REPORT

# Successful Pregnancy in a Patient with Pseudomyxoma Peritonei Arising from Ovarian Mucinous Cystadenocarcinoma Treated with Cisplatin

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**A 24-year-old Japanese woman clinically showing pseudomyxoma peritonei arising from ovarian mucinous cystadenocarcinoma FIGO stage Ic is reported. She received intra-abdominal administrations of cisplatin five times following left oophorectomy. After being free of disease for 6 months, she conceived and carried two pregnancies to successful deliveries at 34 and 37 weeks, respectively. At the Cesarean sections, there were no abnormal findings except for right ovarian mucinous cystadenoma. She has had no evidence of recurrence by the time of the 60-month postoperative examination.** © 1995 Academic Press, Inc.

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### INTRODUCTION

Pseudomyxoma peritonei (PP) is a rare condition found in less than 1 in 10,000 surgical procedures [1] or 3 in 8100 autopsies [2]. This clinical entity is defined by the presence of gelatinous ascites and implants of the tumor on the peritoneal surface and omentum. However, visceral invasion is uncommon.

PP occurs most commonly in mucinous tumors of the ovary and appendix, especially of well-differentiated [3] or borderline malignancy types [4]. The multiple recurrences of intra-abdominal gelatinous ascites may lead to eventual death from inanition [5].

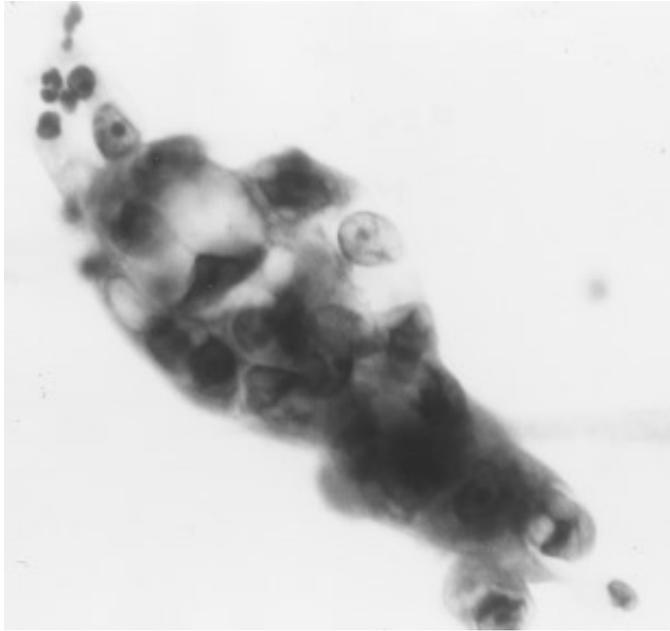
This report describes a case of PP originating from ovarian mucinous cystadenocarcinoma, FIGO stage Ic. Intra-abdominal administrations of cisplatin induced freedom from disease, and the patient carried two successful pregnancies with viable infants.

### CASE REPORT

A 24-year-old Japanese woman, complaining of a full abdominal sensation, consulted a local physician in August 1989. On August 31, paracentesis of the massive ascites was performed and cytological examination showed mucinous cystadenocarcinoma (Fig. 1). The patient was then referred to the Department of Obstetrics and Gynecology, Gifu University Hospital, for further management. She had experienced amenorrhea for the past 6 months. Past history and general physical examinations were unremarkable except for the distended abdomen. Pelvic examination showed an enlarged mass, and transabdominal ultrasonography revealed a multilobular ovarian tumor of approximately 20 × 15 cm. The tumor markers were positive [CA125, 54 U/ml (<35); CA19-9, 1120 U/ml (<37); sialyl Lewis<sup>x-i</sup> antigen, 55.5 U/ml (<38)].

On September 14, 1989, she was submitted to exploratory laparotomy on the working diagnosis of ovarian carcinoma. At the laparotomy, gelatinous massive ascites (2100 ml) were vacuumed; the cytological finding was confirmed as the same as that described on August 31. The left ovary was found to be replaced by an irregular and cystic tumor, measuring approximately 20 × 15 × 10 cm. The cut surface of the resected tumor appeared multilobular, and a mucin-producing tumor was confirmed. The resected tumor pathologically revealed a mucinous cystadenocarcinoma due to the presence of the apparent stromal invasion (Fig. 2). However, the counterpart ovary, uterus, omentum, stomach, and liver were normal in appearance. An omentectomy was not performed; however, the biopsies and imprint cytology from these portions at the omentum, peritoneum, and other pelvic organs, and lymph node sampling, suspected of being neoplastic, also were found to be negative. Around the small

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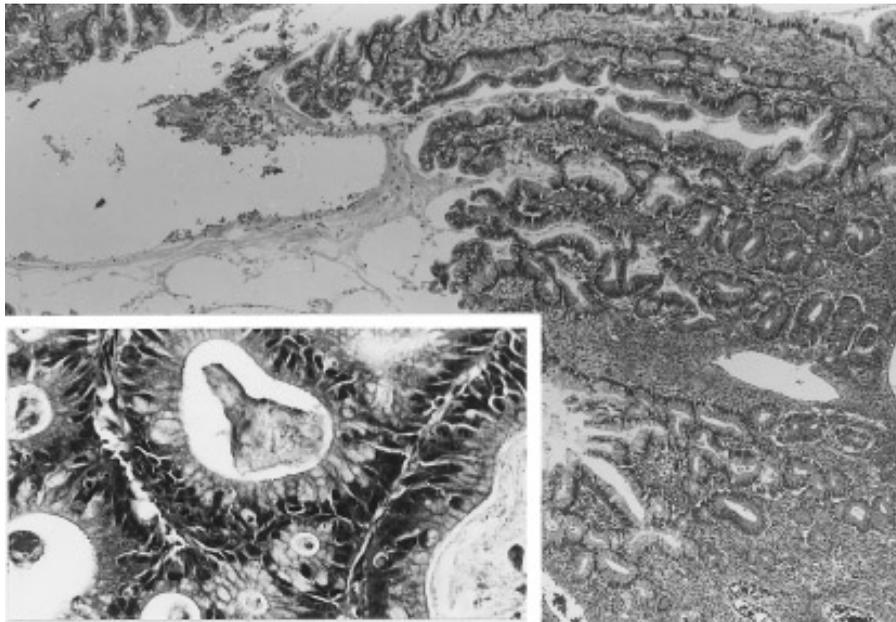


**FIG. 1.** Cytological specimens obtained preoperatively from the ascites. Note crowded clump of malignant cells. Abundant cytoplasm and prominent nucleoli are observed (Papanicolaou stain,  $\times 700$ ).

intestine, gelatinous ascites were found, suggesting the diagnosis of PP. Although PP is sometimes considered to arise from the appendix, the resected appendix showed no abnormalities. Thus, the surgical stage was diagnosed as mucinous cystadenocarcinoma of the left ovary, FIGO stage Ic with PP.

At closing of the abdomen, cisplatin ( $70 \text{ mg/m}^2$ ; body

surface,  $1.43 \text{ m}^2$ ) was administered into the abdominal cavity and a transabdominal indwelling tube was placed for postoperative adjuvant chemotherapy. Postoperative recovery was completely uneventful, and the patient received additional chemotherapies (intra-abdominal administration of cisplatin,  $70 \text{ mg/m}^2$ ; body surface,  $1.43\text{--}1.44 \text{ m}^2$ ) on September 29, October 16, October 30, and November 22, 1989; total



**FIG. 2.** Pathological specimens obtained at the first surgery, diagnosed as mucinous cystadenocarcinoma. Large pools of mucin occupy the ovarian stroma. Stromal invasions are seen. H & E,  $\times 85$ . Inset: "Back-to-back crowding" and epithelial stratification are found. H & E,  $\times 225$ .

amount of cisplatin, 350 mg/m<sup>2</sup> (body surface, 1.43 m<sup>2</sup>). She was discharged on the 30th postoperative day; at that time her menstrual cycle was recovered. She was followed-up at the outpatient clinic of the department.

After chemotherapy, tumor markers and computed tomography for 2 months showed no abnormalities. The patient married and became pregnant, her last menstrual period starting on November 15, 1990 and lasting 5 days. Premature rupture of membrane (PROM) and breech presentation were noted on July 12, 1991. On July 14, 1991 (34 weeks of gestation), she underwent a Cesarean section (C/S) and was delivered of a healthy boy, weighing 2228 g. During the surgery, cystectomy of a right ovarian cyst, measuring 3 cm in diameter, was also performed. Microscopic examination of the cyst proved it to be a benign mucinous cystadenoma. Pathological examinations of the uterine and omental lesions suspected of being neoplastic and imprint cytological examinations of other pelvic organs showed no evidence of neoplasia.

The patient became pregnant again and on June 14, 1993 (37 weeks of gestation), she was delivered of another healthy boy, weighing 3006 g, through a C/S due to PROM. On the second C/S, there were no detectable (pre)neoplastic abnormalities in the abdominal cavity. There was no evidence of disease on follow-up with physical examinations, chest radiographs, and serial ultrasonic evaluations or magnetic resonance imaging for 60 months.

## DISCUSSION

To our knowledge, this is the first report of a successful pregnancy of a patient with PP treated with intra-abdominal administration of cisplatin. In the case presented here, repeated intra-abdominal cisplatin administrations were effective in the treatment of PP arising from ovarian well-differentiated mucinous cystadenocarcinoma.

The diagnosis of PP is made clinically when gelatinous mucin is found filling the abdominal cavity. The histogenesis of this entity remains unclear; simple rupture of a mucinous lesion is not sufficient to explain the cause of PP [6, 7]. In this case, an apparent rupture of the ovarian tumor was not observed at surgery.

PP carries a poor prognosis, even when originating from benign diseases. The treatment of PP is primarily surgical. Oophorectomy, appendectomy, and aggressive debulking of the intra-abdominal tumor have resulted in long-term survival [8]. There have been no reports of long-term survival

of patients treated with an adjuvant therapy, multiple systemic and intra-abdominal chemotherapeutic agents, and less extensive surgery. Cisplatin-based regimens have become the standard for treating ovarian malignancy, and a few groups have reported effective treatment of PP with cisplatin [9, 10]. Here, we presented the first report of a good-outcome pregnancy following cisplatin treatment. In the cisplatin-responsive cases, including ours, the diseases were limited to the well-differentiated or low potential of malignancy types.

Generally, patients with malignant ovarian tumors are necessarily treated with surgical excision. However, in the case of PP, it is difficult to achieve satisfactory reduction through surgery. Although this report is preliminary, the dismal prognosis of the disease prompted us to report the treatment with intra-abdominal administration of cisplatin in the PP case for the conservation of female reproduction.

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## REFERENCES

1. Campbell, J. S., Lou, P., Ferguson, J. P., Krongold, I., Kemeny, T., Mitton, D. M., and Allan, N. Pseudomyxoma peritonei et ovarii with occult neoplasms of appendix, *Obstet. Gynecol.* **42**, 897–902 (1973).
2. Rosenfeld, E. D. Peritoneal pseudomyxoma: Four unusual cases, *Arch. Pathol.* **48**, 255–273 (1949).
3. Cariker, M., and Dockerty, M. Mucinous cystadenomas and mucinous cystadenocarcinomas of the ovary: A clinical and pathological study of 355 cases, *Cancer* **7**, 302–310 (1954).
4. Kaern, J., Trope, C. G., and Abeler, V. M. A retrospective study of 370 borderline tumors of the ovary treated at the Norwegian Radium Hospital from 1970 to 1982: A review of clinicopathologic features and treatment modalities, *Cancer* **71**, 1810–1820 (1993).
5. Mann, W. J., Jr., Wagner, J., Chumas, J., and Chalas, E. The management of pseudomyxoma peritonei, *Cancer* **66**, 1636–1640 (1990).
6. Jones, D. H. Pseudomyxoma peritonei, *Br. J. Clin. Pract.* **19**, 675–679 (1965).
7. Hart, W. R., and Norris, H. J. Borderline and malignant tumors of the ovary: Histologic criteria and clinical behavior, *Cancer* **31**, 1031–1045 (1973).
8. Parsons, J., Gray, G. F., and Thorbjarnarson, B. Pseudomyxoma peritonei. *Arch. Surg.* **101**, 545–549 (1970).
9. Michael, H., Sutton, G., and Roth, L. Ovarian carcinoma with extracellular mucin production: Reassessment of "pseudomyxoma ovarii et peritonei," *Int. J. Gynecol. Pathol.* **6**, 298–312 (1987).
10. Jones, C. M., and Homesley, H. D. Successful treatment of pseudomyxoma peritonei of ovarian origin with cisplatin, doxorubicin and cyclophosphamide, *Gynecol. Oncol.* **22**, 257–259 (1985).