

Serous papillary cystic ovarian borderline tumor: Case report

R. Idotta¹, M.D.; P. Scopelliti², M.D.; C. Polimeni³, M.D.; M. F. Lirosi³, M.D.

¹Gynecologic Department 2° - Casa di cura "Villa Aurora" - Reggio Calabria (Italy)

Summary

A case of a 30-year-old patient with a serous papillary borderline tumor of the right ovary who underwent a simple mono-lateral salpingo-oophorectomy is presented. Conservative surgery allowed a subsequent pregnancy with spontaneous delivery 27 months later.

Key words: Serous-papillary cystic adenoma of the ovary; Borderline; Endoscopic surgery.

Introduction

It is known that borderline ovarian tumors have a good prognosis to the point that the last few years it has allowed non-mutilating interventions in young women who wish to preserve their fertility [1-8]. The new surgical orientation in daily practice is to preserve fertility during surgical therapy of low potential malignant tumors [9-20]. Often these borderline tumors strike patients in fertile age because they tend to develop even ten years before ovarian tumors. Stage I tumors have a good prognosis and include 75-85% of cases [21]. Recurrences after conservative therapy to maintain fertility are greater after ovarian resection for cysts (12-37%) [22] than after mono-lateral salpingo-oophorectomy (0-22%). Spontaneous pregnancies after conservative surgery are rare (15%). Parker and co-workers found no differences in survival rates at five years both for patients treated with hysterectomy and salpingo-oophorectomy and for those treated only with mono-lateral salpingo-oophorectomy [23]. The latter may be the definitive treatment for young patients with low parity and well-differentiated serous carcinoma [24, 25].

The tumor must be mono-lateral, well capsulated, free from adhesions and without ascites or evident extra-gonadal dissemination. Peritoneal washings for cytology must be negative [26-28]. Scrupulous evaluation of the contralateral ovary must be carried out together with close follow-up [29-33].

We describe a case which confirms such clinical procedure in the world cases.

Case report

The patient, age 30, came under our care on December 16, 1999 following a right adnexal tumor of an unknown type. Such neoplasm appeared ecographically pluriloculated with cystic proliferations and hypervascularized walls (Figure 1). Additionally it showed endophytic proliferation at the superior pole.

Therefore a laparoscopy was performed which confirmed the echographic results. Considering the risk of peritoneal dissemination related to endoscopic surgery and the evaluation of the ovarian malignancy, we proceeded prudently with a mini-laparotomy and right salpingo-oophorectomy.

Together with the removed surgical material, 4 cc of free fluid in the peritoneum was sent for examination. The histologic exam revealed a serous papillary cystic tumor of the ovary with a low degree of malignancy (borderline). Considering the young age of the patient and her wish to have children, we decided to preserve the uterus and the contralateral adnexa which were however macroscopically normal.

The definitive histological exam of the surgical materials was: borderline serous papillary tumor with multiple capsular micro-invasion and tuba without lesions. The search for cell tumoral malignancy (CTM) in the peritoneal liquid was negative (Figure 2).

Histology:

A multilocular cystic tumor was present in the ovary. The cysts, often filled by serous fluid material, presented fibrous walls and multiple broad papillae, markedly hypocellular, and edematous tissue projecting into the cysts.

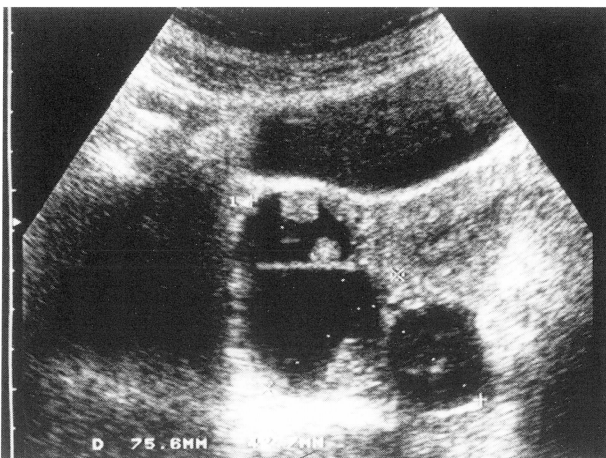


Figure 1. — Ultrasound image of the right ovarian pleuricystic structures with endophytic proliferation.

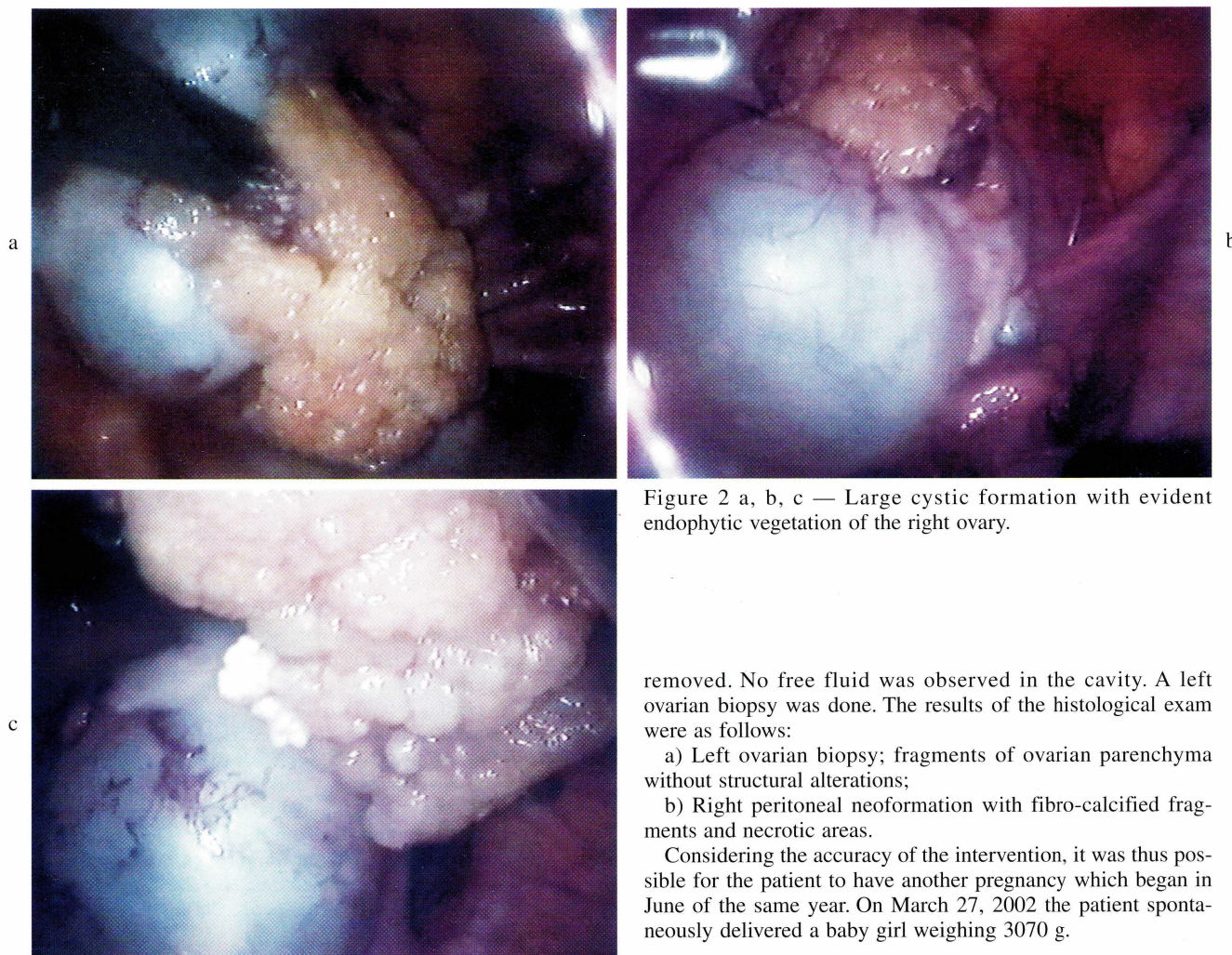


Figure 2 a, b, c — Large cystic formation with evident endophytic vegetation of the right ovary.

The epithelium lining the cysts as well as the papillae was serous type, unilayered, lacking nuclear atypia.

However in the stromal papillae multiple foci (>20% of the surface) of epithelial proliferation, with cellular stratification and small papillary cluster detachment within the cysts could be seen.

The epithelium displayed different features: diffuses ciliated cells, focally rounded mesothelial-like cells with prominent microvillous differentiation of the cytoplasmic border, and hobnail-like cells could be observed.

Only very few scattered mitoses were present in the basal layer.

No stromal invasion nor medusa-like epithelial proliferation was found.

The diagnosis was of a serous papillary cystoadenofibroma borderline or so-called atypical proliferation.

The remnant of the ovarian cortex displayed maturing follicles and a few small follicle cysts.

The Fallopian tube was normal.

After the intervention with mono-lateral salpingo-oophorectomy, combined extra-progestins were given for six months. The tumor markers, echography and clinical controls were always negative. After one year (January 22, 2001) a second-look laparoscopy was done which showed a small whitish formation near the round right ligament; such formation was

removed. No free fluid was observed in the cavity. A left ovarian biopsy was done. The results of the histological exam were as follows:

- a) Left ovarian biopsy; fragments of ovarian parenchyma without structural alterations;
- b) Right peritoneal neoformation with fibro-calcified fragments and necrotic areas.

Considering the accuracy of the intervention, it was thus possible for the patient to have another pregnancy which began in June of the same year. On March 27, 2002 the patient spontaneously delivered a baby girl weighing 3070 g.

Discussion

The present case shows – in agreement with the world literature – how young fertile women who potentially desire children can undergo conservative treatment for borderline ovarian tumors. This solution needs to be part of the competence of the gynecologist in the therapeutic approach [16, 21, 26], considering that the rationality of such conservative attitude is based on three points:

- 1) To preserve fertility;
- 2) To preserve endocrine activity;
- 3) To safeguard the “self-image” of the woman [9-13, 18-21].

It is clear that the possibility of conservative surgery is conditioned by stage, mono- or bilaterality and the histological characteristics of the neoplasia [13-17]. Mucinous and endometrioid lesions have a better prognosis than serous lesions, and Stage I and borderline lesions are easier to treat with conservative treatment. Serous lesion [33] show a frequency of bilaterality seven times higher with respect to mucinous carcinomas. After a surgical procedure to save tissue, in cases in which chemotherapy is proposed, it could destroy the remaining oocytes, especially in patients over age 30.

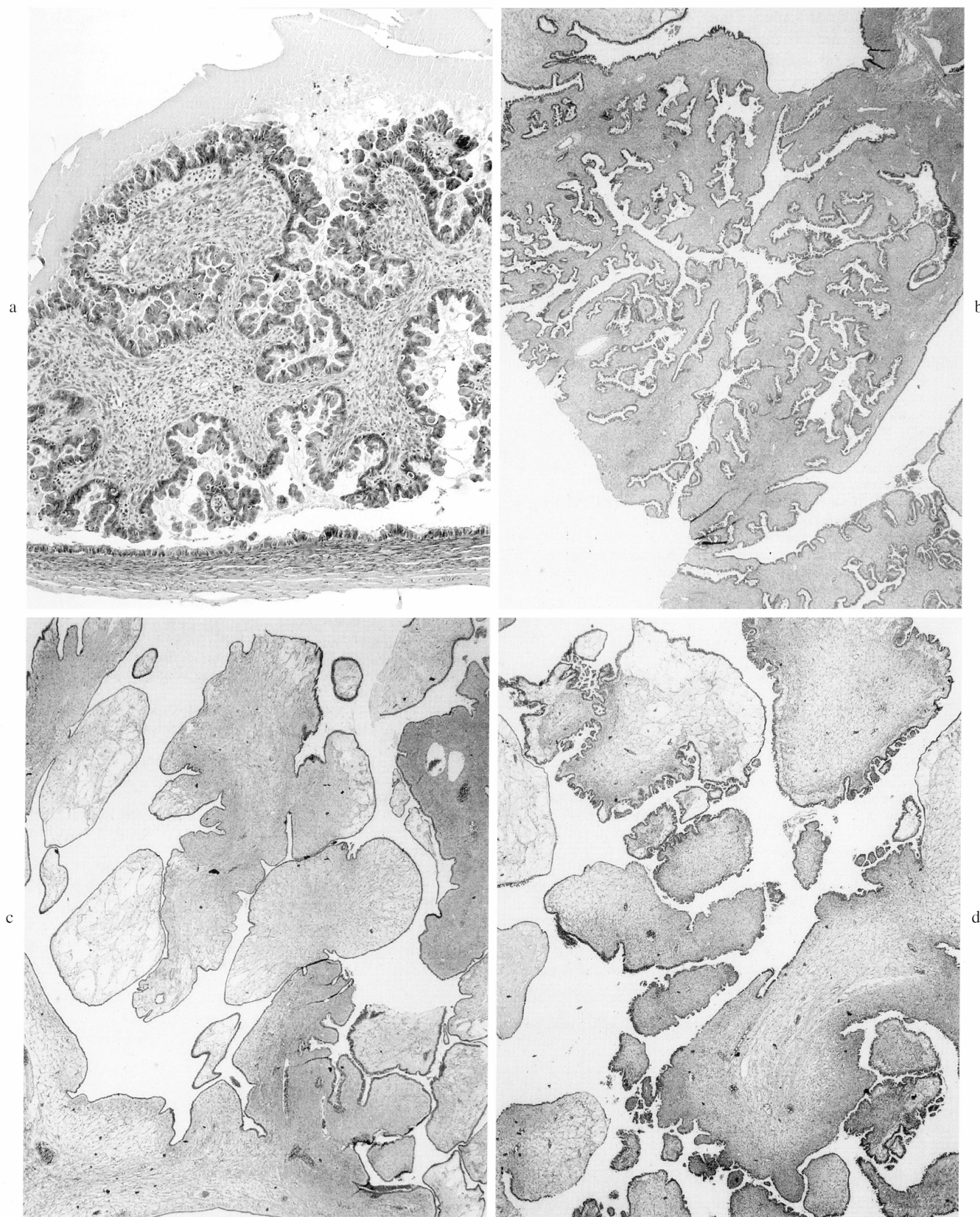


Figure 3. — a) Endocystic papillary serous mucinous tumor with epithelial proliferation and detachment of cellular cluster (x 12). b) Papillary serous adenofibroma with broad edematous stroma (x 3). c) Papillary serous adenofibroma with and low grade epithelial proliferation. d) Papillary serous adenofibroma with scattered foci of low grade epithelial proliferation (x 3).

In Stage I ovarian cancer cases, it has been shown that young women aged between 16 and 29 who are treated with mono-lateral salpingo-oophorectomy or only with resection of the ovary do not have recurrences in a follow-up period of three to ten years [9-13, 16, 21, 22].

This case is included in the about 10-15% of epithelial ovarian tumors are represented in fact by neoplasias with low malignant potential, called borderline, that frequently arise in women in fertile age [1-8, 21, 22, 27].

References

- [1] D. S. Papadimitriou, P. Martin-Hirsch, H. C. Kitchener, D. E. Lolis, N. Dalkalitsis, E. Paraskevaidis: "Recurrent borderline ovarian tumours after conservative management in women wishing to retain fertility". *European Journal of Gynaecol. Oncol.*, 1999, 2, 94.
- [2] R. Halperin, S. Zehavi, P. Dar, L. Habler, E. Hadas, I. Bukovsky, D. Schneider: "Clinical and molecular comparison between borderline serous ovarian tumors and advanced serous papillary ovarian carcinomas". *European Journal of Gynaecol. Oncol.*, 2001, 4, 292.
- [3] A. Basta, K. Pitynski, W. Krysztopowicz, D. Przeszlakowski: "Epithelial ovarian tumors of borderline malignancy". *European Journal of Gynaecol. Oncol.*, 1999, suppl. 89.
- [4] J. Prat: "Serous borderline tumors of the ovary". *European Journal of Gynaecol. Oncol.*, 2001, suppl. 11.
- [5] H. Makarewicz, J. Emerich, J. Olszewski, J. Kobierski, B. Krolkowska: "Epithelial ovarian tumors of borderline malignancy: A follow up study of 127 cases". *European Journal of Gynaecol. Oncol.*, 2001, suppl. 70.
- [6] P. Morice, S. Camatte, P. Pautier, C. Lhomme, C. Haie-Meder, P. Duvillard, D. Castaigne: "Conservative treatment of ovarian borderline tumor". *European Journal of Gynaecol. Oncol.*, 2001, suppl. 211.
- [7] A. Uppin, I. Vaasna, L. Kibe, V. Padrik, H. Saar: "Ovarian tumours of borderline malignancy – our experience". *European Journal of Gynaecol. Oncol.*, 2001, suppl. 213.
- [8] P. Kclavic, G. Grubiasic, L. Bolf Benkovic, A. Pirkic: "Advanced ovarian borderline tumor". *European Journal of Gynaecol. Oncol.*, 2001, suppl. 228.
- [9] Wu P. C., Huang R. L., Lang J. H., Lian L. J., Tang M. Y.: "Treatment of malignant ovarian germ cells tumors with preservation of fertility: a report of 28 cases". *Gynecol. Oncol.*, 1991, 40, 2.
- [10] Miyazaki T., Tomoda Y., Ohta M. *et al.*: "Preservation of ovarian function and reproductive ability in patients with malignant ovarian tumors". *Gynecol. Oncol.*, 1988, 30 (3), 329.
- [11] Menczer J.: "A trend toward more conservative surgery in gynecologic oncology". *Obstet. Gynecol. Surv.*, 1996, 51 (10), 628.
- [12] Gonzales-Lira G., Escudero-De Los Rios P. *et al.*: "Conservative surgery for ovarian cancer and effect on fertility". *Int. J. Gynecol. Obstet.*, 1997, 56 (2), 155.
- [13] Gotlieb W. H., Flikker S. *et al.*: "Borderline tumors of the ovary: fertility treatment, conservative management and pregnancy outcome". *Cancer*, 1998, 82, 141.
- [14] Prat J.: "Borderline tumors of the ovary". *Atti della Sigo*, 1996, LXXII, 403.
- [15] Linkej S. R., Reede *et al.*: "Borderline ovarian tumors". *Am. S. Med.*, 1996, 101, 217.
- [16] Fontanelli R., Brescianti G.: "Trattamento delle forme 'Borderline'". *Atti della Sigo*, 1996, LXXII, 410.
- [17] Trimble C. L., Trimble E. L.: "Management of epithelial ovarian tumors of low malignant potential". *Gynecol. Oncol.*, 1994, 55, 552.
- [18] Young R. C., Pecorelli S.: "Management of early ovarian cancer". *Semin. Oncol.*, 1998, 25 (3), 335.
- [19] Tserke Zougrou A. J.: "Malignant ovarian neoplasm. The place of conservative surgery". *Ann. N. Y. Acad. Sci.*, 1997, 816, 362.
- [20] Snider D. D., Stuart G. C. E., Nation J. G., Robertson D. I.: "Evaluation of surgical staging in stage I, low malignant potential ovarian tumors". *Gynecol. Oncol.*, 1991, 40, 129.
- [21] Onnis A., Marchetti M., Piazza M.: "Clinical experience in gynecological cancer management". *Eur. J. Gynecol. Oncol.*, 1993, 14, 17.
- [22] Lim-Tansk, Cajigas H. B., Scully R. E.: "Ovarian cystectomy for serous borderline tumors: a follow-up study of 35 cases". *Obstet. Gynecol.*, 1988, 72, 775.
- [23] Parker W. H., Berek J. S.: "Management of selected cystic adnexal masses in postmenopausal women by operative laparoscopy: a pilot study". *Am. J. Obstet. Gynecol.*, 1993, 157, 1574.
- [24] Twaalfhoven F. C. M., Peters A. A. W. *et al.*: "The accuracy of frozen section diagnosis of ovarian tumors". *Gynecol. Oncol.*, 1991, 41, 189.
- [25] Menzin A. W., Rubin S. C. *et al.*: "The accuracy of a frozen section diagnosis of borderline ovarian malignancy". *Gynecol. Oncol.*, 1995, 59, 183.
- [26] Creasman W. T., Park R., Norris H. *et al.*: "Stage borderline ovarian tumors". *Obstet. Gynecol.*, 1982, 59, 93.
- [27] Chambers R. J., Merino M. J., Kohorn E. I., Schwartz T. E.: "Borderline ovarian tumors". *Am. J. Obstet. Gynecol.*, 1988, 159, 1088.
- [28] Bourne T., Campbell S. *et al.*: "Screening for early familial ovarian cancer with transvaginal ultrasonography and colour blood flow imaging". *Br. Med. J.*, 1993, 306, 1025.
- [29] Sassone A. M., Timor-Tritsch I. E., Artnet *et al.*: "Transvaginal sonographic characterization of ovarian disease: evaluation of a new scoring system to predict ovarian malignancy". *Obstet. Gynecol.*, 1991, 78, 70.
- [30] Sulian C. G., Wooiruff J. D.: "The biologic behaviour of low-grade papillary serous carcinoma of the tumors". *Obstet. Gynecol.*, 1972, 40, 860.
- [31] Hopkins M. P., Morley G. W.: "The second-look operation and surgical reexploration in ovarian tumors of low malignant potential". *Obstet. Gynecol.*, 1989, 74, 375.
- [32] Kurjak A., Shalan H., Kupesic S. *et al.*: "Transvaginal color doppler sonography in the assessment of pelvic tumor vascularity ultrasound". *Obstet. Gynecol.*, 1993, 3, 137.
- [33] Gershenson D. M., Silva E. G.: "Serous ovarian tumors of low malignant potential with peritoneal implants". *Cancer*, 1990, 65, 578.

Address reprint requests to:
R. IDOTTA, M.D.
Via G. Melacrino, 80
89124 Reggio Calabria (Italy)