Monodermal highly specialized teratoma of the ovary: a sebaceous gland tumor

C. Cicchini[1], *M.D.*; A. Larcinese[1], *M.D.*; F. Ricci[2], *M.D.*; P. Aurello[1], *M.D.*; P. L. Mingazzini[2], *M.D.*; M. Indinnimeo[1], *M.D.*

[1]Department of Surgery "Pietro Valdoni",
[2]Department of Experimental Medicine and Pathology, University of Rome " La Sapienza", Rome (Italy)

Summary

Teratomas are neoplasms that originate in pluripotential cells and contain representations of all three germ layers in a rather mature state. Specialized forms of teratoma with unilateral development of certain tissues, such as struma ovarii, argentaffin tumors, cholesteatoma, primary choriocarcinoma of the ovary, pseudomucinous cystoma and neurogenic cysts are known. In this paper we describe an ovarian teratoma consisting entirely of sebaceous glands.

Key words: Teratoma; Ovary; Sebaceous gland; Monodermal.

Introduction

Teratomas are neoplasms that originate in pluripotential cells and are composed of a wide diversity of tissue foreign to the organ or anatomic site in which they arise. The tumors frequently occur in young adults, with equal incidence in both sexes. The gonads are the first most frequent location, followed by the anterior mediastinum. Occasionally they occur in the pericardium or posterior mediastinum or other locations [1-9]. The tumors contain representations of all three germ layers in a rather mature state. When the lesions are cystic and contain hair and teeth, they are called dermoid cysts, but this is a misnomer because these tumors are not of ectodermal origin. In 1964 Strauss and Gates first described a sebaceous gland tumor of the ovary almost entirely consisting of sebaceous glands, regarding this tumor as a variant of a dermoid cyst characterized by unilateral development [10]. Another similar case was reported in the literature in the last 34 years [11]. In this paper we describe an ovarian teratoma consisting entirely of sebaceous glands.

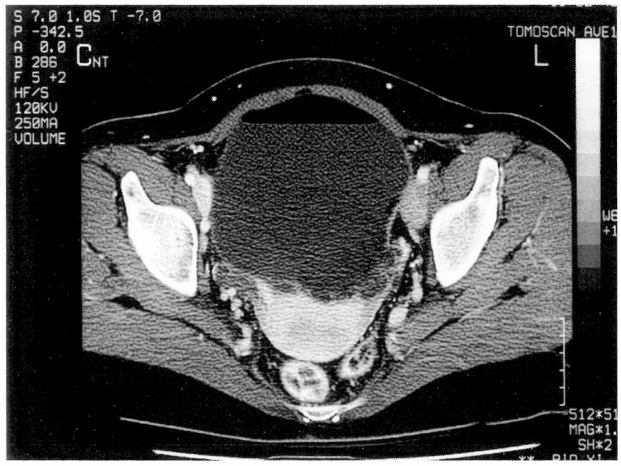

Figure 1. — A voluminous mass (16x13 cm in diameter) in the right inguinal deforming the abdominal wall and the bladder was detected at angio TC. The neoplasm seemed fatfilled and well capsulated, not infiltrating neighbouring organs.

Case Report

A 37-year-old woman, para 2, gravida 3, aborta 1, entered the hospital complaining of painless abdominal swelling of two months' duration. Physical examination revealed a large mass filling the hypogastrium almost up to the umbelicus which was thought to be attached to the left ovary. Her past medical history was non contributory except for tonsillectomy at the age of five. Ultrasonic tomography revealed a cystic neoplasm with septa and mild vascularization at power doppler. The neoplasm was 30 cm in diameter and appeared to be from the right ovary. The bladder and uterus were dislocated. Angio TC detected a voluminous mass (16x13 cm in diameter) in the right inguinal deforming the abdominal wall and the bladder (Figure 1). The neoplasm seemed fatfilled and well capsulated, not infiltrating

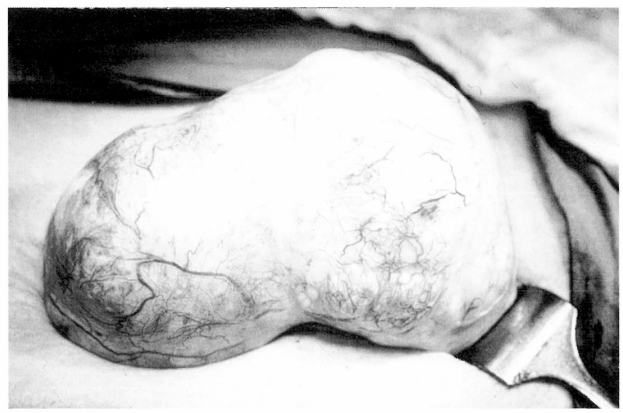

Figure 2. — Macroscopic appearance of the neoplasm at laparotomy.

Revised manuscript accepted for publication February 22, 2002

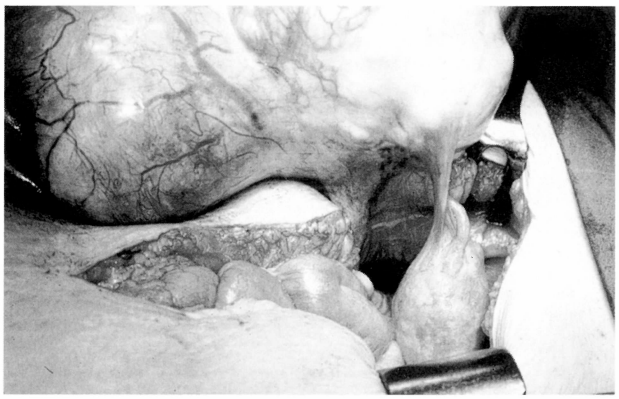

Figure 3. — Neoplasm originating from the left ovary.

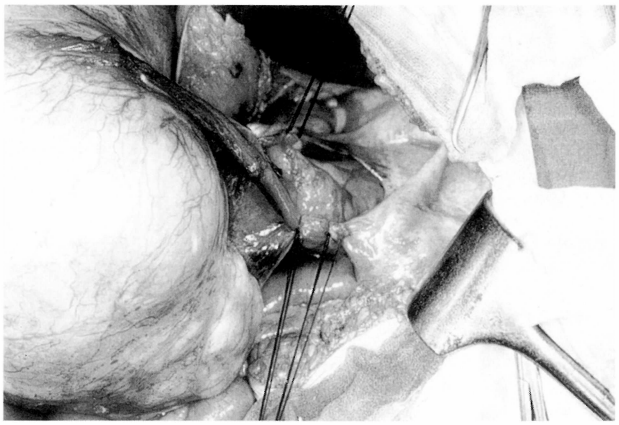

Figure 4. — Left salpingo-oophorectomy was performed.

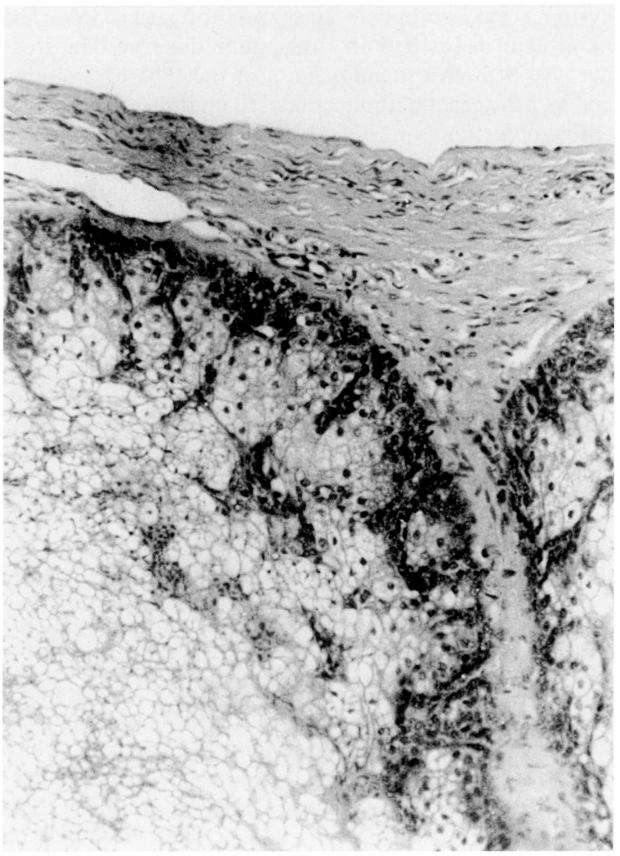

Figure 5. — At microscopic examination the generative cells tended to be arranged at the periphery of the lobules and the second-type cells were lipid-laden mature sebaceous cells occupying the center of the glands. Atypical mitotic figures were absent.

neighbouring organs. Secondary mild hydroureteronephrosis on the right side was associated. Radiographic diagnosis was a dermoid cyst of the left ovary. The right adnexa was normal. No lymphadenopathy or metastatic localization were found. Laboratory data on admission were within normal limits except for CA125 dosage (CA125 43 U/ml, normal range < 35 U/ml). On June 2001 she underwent surgical exploration. Laparotomy revealed a large left ovarian mass that filled the abdomen (Figures 2, 3), and 200 ml of transparent fluid were present in the peritoneal cavity. A left salpingo-oophorectomy was performed (Figure 4). The postoperative course was uneventful. Cytologic examination of the peritoneal fluid was negative for neoplastic cells. She was well six months after surgery. The excised ovarian mass was a unilocular cyst 28 x 12 x 8 cm in diameter weighing 1980 g and filled with sebaceous material. The serosa was intact and the smooth outer surface was glistening white. The consistency of the growth resembled that of dough and it seemed to be yellow as viewed through the serosa. Sectioning revealed that the periphery of the tumor was yellow, soft, fatty and unusually friable toward the center. Masses of papillary-like tissue protruded over the otherwise smooth inner surface of the cyst. The neoplasm consisted of lobules of sebaceous glands which varied in size and formed cystic structures filled with sebum. The lobules included two elements: 1) small basophilic cells, the generative cells of the basal layer of the sebaceous glands, 2) the larger sebaceous cells proper (Figure 5). The generative cells tended to be arranged at the periphery of the lobules and were identical to those present at the periphery of the normal sebaceous glands. The second type of cells were large lipid-laden mature sebaceous cells occupying the center of the glands with round central nuclei. Atypical mitotic figures were absent. Histologic diagnosis was highly specialized sebaceous monodermal teratoma (sebaceous gland tumor of the ovary).

Discussion

Ovarian germ-cell tumors are rare, accounting for about 2% to 3% of all ovarian cancers. These tumors may have a mixed histologic pattern, and treatment should be designed to deal with the most malignant component [12]. A thorough search of the pertinent literature revealed only two previous cases dealing with a solid neoplasm composed almost entirely of sebaceous glands [10, 11]. We regard these tumors as teratogenous neoplasms originating from unilateral development of sebaceous glands of a dermoid. This tumor histologically resembles a sebaceous adenoma of the skin in an extraocular location and shows benign behavior. Specialized forms of teratoma with unilateral development of certain tissues, such as struma ovarii, argentaffin tumors, cholesteatoma, primary choriocarcinoma of the ovary, pseudomucinous

cystoma and neurogenic cysts, are known [13-20]. The neoplasm described in this paper deserved interest because of both the uniqueness of the type of teratoma and its histogenetic implications. Even if there is no theoretical objection to the origin of monodermal tumor from the cells of sebaceous units, it is more usual for sebaceous elements to be differentiated from the cells of carcinoma of squamous cell character [21, 22]. This occurrence was excluded in our case in view of the fact that squamous epithelium showed no abnormalities at all. Why a teratoma should develop in a one-sided direction is not known. Growth of sebaceous glands is determined by genetic influence and sebaceous glands are target organs of internal secretory influences. The patient described in this paper had no clinical signs of an internal secretory abnormality. Evocators or evocator-like chemical stimulation may have a role in the unilateral purely sebaceous development of the tumor. Breakdown products of sebaceous secretions, such as oleic acid, may make the cells more sensitive to stimulating influences, i.e., hormones or chronic irritation, and may even be a factor influencing malignant change. In evaluating the sebaceous tumor of the ovary described in the paper, the ovarian growth had a well-defined capsule as the benign cutaneous tumor. In contra-distinction to sebaceous tumors of the skin, the ovarian growth arose on a teratogenetic basis and the size of a neoplasm is generally not a criterion of malignancy, especially in the ovary. The lesion in the present paper was sharply demarcated and composed of mature and immature sebaceous gland cells arranged in an organoid pattern. Even if a sebaceous adenoma may suffer malignant change we could not claim this occurrence in our case because no adenomatous tissue could be found.

References

[1] Cortes J., Llompart L., Rossello J. J., Rifa J., Mas J., Anglada P., *et al.*: "Immature teratoma primary of the uterine cervix. First case report". *Eur. J. Gynaecol. Oncol.*, 1990, *11*, 37.

[2] Beutel K., Partsch C. J., Janig U., Nikischin W., Suttorp M.: "Oral mature teratoma containing epididymal tissue in a female neonate". *Lancet.*, 2001, *357*, 283.

[3] Otani M., Tsujimoto S., Miura M., Nagashima Y.: "Intrarenal mature cystic teratoma associated with renal dysplasia: case report and literature review". *Pathol. Int.*, 2001, *51*, 560.

[4] Jao M., Kang Y. K., Lee H. K., Lee H. S., Yum H. K., Bang S. W., *et al.*: "Intrapulmonary and gastric teratoma: report of two cases". *J. Korean Med. Sci.*, 1999, *14*, 330.

[5] Goyal M., Sharma R., Sawhney P., Sharma M. C., Berry M.: "The unusual imaging appearance of primary retroperitoneal teratoma: report of a case". *Surg. Today*, 1997, *27*, 282.

[6] Sakurai Y., Uraguchi T., Imazu H., Hasegawa S., Matsubara T., Ochiai M. *et al.*: "Submucosal dermoid cyst of the rectum: report of a case". *Surg. Today*, 2000, *30*, 195.

[7] Kurosaki Y., Tanaka Y. O., Itai Y.: "Mature teratoma of the posterior mediastinum". *Eur. Radiol.*, 1998, *8*, 100.

[8] Sepulveda W., Gomez E., Gutierrez J.: "Intrapericardial teratoma". *Ultrasound Obstet. Gynecol.*, 2000, *15*, 547.

[9] Yoshioka T., Tanaka T.: "Mature solid teratoma of the fallopian tube: case report". *Eur. J. Obstet. Gynecol. Reprod. Biol.*, 2000, *89*, 205.

[10] Strauss A. F., Gates H. S.: "Giant sebaceous gland tumor of the ovary". *Am. J. Clin. Pathol.*, 1964, *41*, 78.

[11] Kaku T., Toyoshima S., Hachisuga T., Enjoji M., Tanaka M.: "Sebaceous gland tumor of the ovary". *Gynecol. Oncol.*, 1987, *26*, 398.

[12] Hirai K., Ishiko O., Itoh F., Nakagawa E., Kanaoka Y., Sumi T. *et al.*: "Clinical evaluation of mature teratomas containing malignant elements". *Oncol. Rep.*, 2000, *7*, 655.

[13] Urbano U., Facchini V., Gadducci A., Colombi L., Prato B.: "Struma ovarii. Observations on three cases". *Eur. J. Gynaecol. Oncol.*, 1984, *5*, 113.

[14] Ferrer J., Herrero-Zapatero A., Orille V., Llaneza P., Perez-Rodrigo C.: "Thyroid carcinoma originating in a teratoma of the ovary". *Eur. J. Gynaecol. Oncol.*, 1995, *16*, 396.

[15] Esik O., Nemeth G., Szepeshazi K.: "Calcitonin-secreting ovarian strumal carcinoid". *Eur. J. Gynaecol. Oncol.*, 1994, *15*, 211.

[16] Miliauskas J. R.: "Carcinoid tumor occurring in a mature testicular teratoma". *Pathology*, 1991, *23*, 72.

[17] Roncaroli F., Scheithauer B. W., Pires M. M., Rodrigues A. S., Pereira J. R.: "Mature teratoma of the middle ear". *Otol. Neurotol.*, 2001, *22*, 76.

[18] Zahn C. M., Kendall B. S.: "Heterotopic bone in the ovary associated with a mucinous cystoadenoma". *Mil. Med.*, 2001, *166*, 915.

[19] Jain T., VanKessel K., Reed S., Paley P.: "Leydig cell tumor, mature teratoma, and nongestational choriocarcinoma in a single ovary". *Obstet. Gynecol.*, 2000, *95*, 1031.

[20] Sah S. P., Verma K., Rani S.: "Neurogenic cyst of ovary: an unusual massive monodermal teratoma". *J. Obstet. Gynaecol. Res.*, 2001, *27*, 21.

[21] Kontogianni E., Koukoura E., Christopoulou E.: "Squamous cell carcinoma arising in mature cystic teratoma of the ovary: a case report". *Eur. J. Gynaecol. Oncol.*, 2001, *22*, 238.

[22] Bontis J., Vakiani M., Vavilis D., Agorastos T., Dragoumis K.: "Squamous cell carcinoma arising from mature cystic teratoma: a report of three cases". *Eur. J. Gynaecol. Oncol.*, 1996, *17*, 49.

Address reprint requests to:
C. CICCHINI, M.D.
Via Cairano, 6
00177 Rome (Italy)