Case report

Xanthogranuloma of the vagina

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Xanthogranuloma is an unusual tumor that affects the ovary, mesentery, anal wall, kidney, and retroperitoneum.[1–5] To our knowledge, only 19 cases have been reported.

We report a case of xanthogranuloma of the rectovaginal septum with presenting symptoms of paravaginal tumor. The patient was treated with radiation and surgery.

Case report

A 30-year-old woman, gravida 2, para 2, was referred to the Cleveland Clinic for a large paravaginal mass, which had increased in size in the previous 6 months. Because of the mass, intercourse was difficult. The patient had the mass for 7 years. It had first been noticed during her second pregnancy. A low segment cesarean section had been performed for soft tissue dystocia because the size of the mass had increased. At that time a tubal ligation was also done.

Based on pathologic findings the diagnosis was xanthogranuloma. The lesion regressed after delivery. The local gynecologist had examined her regularly for the past 4 years and had noted an increase in the mass in the 6 months preceding referral to the Cleveland Clinic. Her husband was unable to achieve deep penetration during intercourse.

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Examination at the Cleveland Clinic in June 1977 revealed a 30-year-old woman in no acute distress. On pelvic examination, a firm, rubbery mass about 13 x 10 cm was palpated in the right paravaginal area through the anterior and posterior walls of the vagina. The vaginal mucous membrane overlying the mass was mobile and nonulcerated. The top of the mass could not be reached and the lesion compressed the vagina to a slitlike cavity. The major portion of the mass was in the true pelvis. The cervix was seen only partially above the mass, and the mass was easily palpable in the perineum medial to the right ischial tuberosity. It appeared fixed to the ischial tuberosity and the uterus and adnexa seemed normal.

Palpation through the rectum revealed the same large lesion. The rectal wall appeared fixed to the mass, but the mucous membrane was free. Proctosigmoidoscopy to 25 cm was normal. The scans of the pelvis are shown in *Figures 1 and 2*.

A urogram showed no abnormality. The blood cholesterol level was 285 mg/dl. Examination and cystoscopy were performed under anesthesia. The mucous membrane was edematous at the base of the bladder and the hard tumor was again palpated. Biopsy specimens from the mucous membrane showed chronic trigonitis. Biopsies were obtained transvaginally and transperineally; the tissue was grayish yellow. Histologic diagnosis was xanthogranuloma. Dilatation and curettage of the uterus was also performed at this time. The uterus was enlarged to 10 cm, but no other abnormality was detected. Diagnosis of xanthogranuloma of the vaginal wall with extension into subepithelial planes and into the skeletal muscle was confirmed (*Fig. 3*).

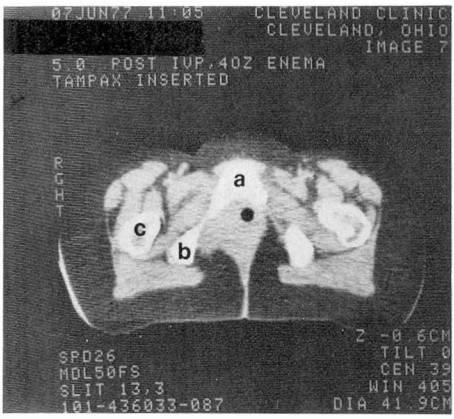

Fig. 1. CT scan of the pelvis with the patient supine. Tampax (black circle) was inserted in the vagina. The mass extends in the right paravaginal area medial to the right ischial tuberosity; a = symphysis pubis, b = ischial tuberosity, c = femur head.

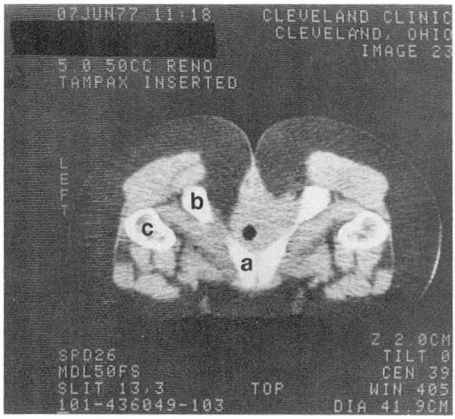

Fig. 2. CT scan of the pelvis with the patient prone. Tampax is shown in the vagina. The mass can be seen in the right paravaginal area between the Tampax and the right ischial tuberosity; a = symphysis pubis, b = ischial tuberosity, c = femur head.

Because of the increasing size of the tumor and functional disability associated with it, 3000 rads were delivered through 17 x 17 cm anterior and posterior pelvic parts daily with a 10 MEV linear accelerator. There was considerable regression of the tumor in nine treatments. Radiation therapy was continued for 15 treatments fractionated at 200 rads/day. Examination at the completion of radiation showed only some thickening in the posterior rectovaginal septum—the mass had disappeared. There were minimal radiation changes of the vagina and the uterus had become smaller. The patient was amenorrheic due to castration by radiation.

To prevent recurrence, it was decided that the rectovaginal septal thickening must be excised. After completion of radiation therapy, a laparotomy was performed through

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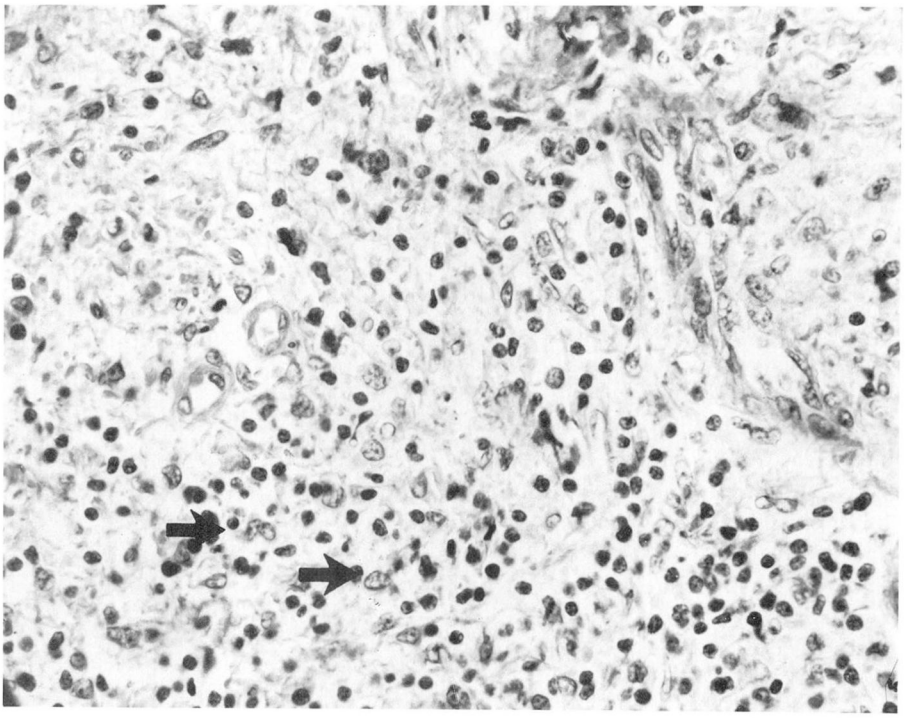

Fig. 3. Biopsy specimen of vagina. A mixed inflammatory infiltrate composed of lymphocytes and histiocytes (arrows) is present in loose connective tissue. Histiocytes contain indistinct cytoplasmic borders and have granular cytoplasm (hematoxylin and eosin stain, ×400).

an old midsubumbilical incision. Examination of the lateral paravaginal region showed no evidence of tumor. The only abnormalities were minimal radiation changes. A total abdominal hysterectomy and bilateral salpingoophorectomy was performed. The patient was transferred to the lithotomy position. An incision was made medial to the right labium minus, through which a fibrotic mass (5 x 2 x 1.5 cm) was removed from the rectovaginal septum (*Fig. 4*). The cavity was packed with gauze and the gauze was removed on the second postoperative day. Healing occurred by secondary intention.

Postoperative examination 6 weeks later showed the vagina to be normal. The patient was given 0.625 mg of Premarin cyclically for control of hot flashes.

Pathologic examination of the organs and the fibrotic mass did not reveal any residual or recurrent xanthogranuloma.

Two years after treatment, there is no evidence of recurrence. The cholesterol level continues to be normal and there is no evidence of lipid or cholesterol abnormality.

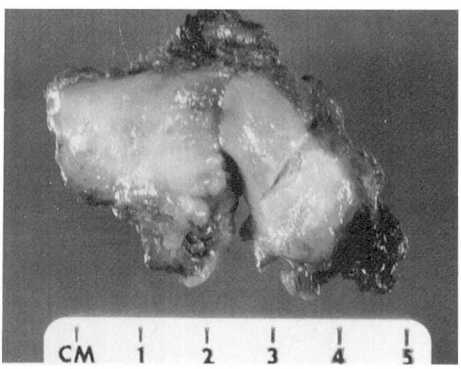

Fig. 4. Scarified remains of the xanthogranuloma removed from rectovaginal septum.

Discussion

Oberling[6] called attention to xanthogranulomas as rare tumors probably related to the "histiocytosis" group of ill-

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nesses, which includes Hand-Schuller-Christian disease, Letterer-Siwe disease, and eosinophilic granuloma of the bone. The histologic characteristics of xanthogranuloma entail fibrosis and chronic inflammation and an abundance of macrophages, lymphocytes, and plasma cells. On light electron microscopic examination, Papadimitriou and Matz[7] described it as myoangiomata and stressed its neoplastic characteristics.

The cause of xanthogranuloma is unknown. Gup,[1] in describing xanthogranuloma, proposed that the embryonic potentials of the ubiquitous connective tissue cells can be awakened by adequate stimulation. The rectovaginal area in a woman exposed to repeated trauma of childbirth and coitus would be a logical site for such development.

Xanthogranulomas are usually solitary but may be multiple. They infiltrate adjacent organs; in our patient the perineum, right paravaginal region, and rectum were invaded by the tumor that arose in the rectovaginal septum. Although xanthogranulomas are usually benign, metastases have been described; the lung has been the common site of metastasis.[8]

The treatment of xanthogranuloma requires individualization. Excision and radiation can be used alone or in combination and vary with the site, extent, and symptoms. In this case, the dramatic response to radiation altered the plan of treatment. An extremely difficult dissection was converted to simple excision of the scarified xanthogranuloma.

In view of the rarity of this tumor, it is important that individual cases be reported to increase the available data and give more insight into the management of these lesions.

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