CASE REPORTS

LEIOMYOMA OF THE OVARY: REPORT OF AN UNUSUAL CASE AND REVIEW OF THE LITERATURE

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LEIOMYOMA is one of the rarest of solid tumours of the ovary. Most authors of current American pathology texts do not mention it, and only two[5, 11] provide more than one sentence on this entity. It therefore appears justified to report an additional and rather unusual case of ovarian leiomyoma, and to focus some attention on this type of neoplasm.

The patient was a 60-year-old white woman, gravida x, para x, whose menopause had occurred at the age of 54. She had one episode of vaginal spotting for five days in September 1959; no treatment was instituted. For ten months before admission, the patient's abdomen became progressively distended and felt very firm and "full", and she experienced occasional bouts of crampy pain in the lower abdomen.

Physical examination showed an asthenic woman with abdominal distress. Her blood pressure was 160/85 mm. Hg, pulse rate 82/min. and temperature 98.6° F. Her chest was clear. Liver, spleen and kidneys were not palpable. A large mass was felt in the lower abdomen which was tympanitic, mobile, smooth and very firm. Preoperatively, the diagnosis of pseudomucinous cystadenoma of the ovary was made.

Laparotomy was performed on October 6, 1960 (by Dr. L. O. Watt).

Pathological findings, gross.—The subtotally resected uterus and both adnexa were submitted for examination. The right ovary was completely replaced by a large, somewhat lobulated cystic structure measuring 26 x 25 x 14 cm. It extended into the mesovarium and into the upper portion of the broad ligament. The right Fallopian tube occupied its usual position, touching the surface of the tumour. No trace of normal ovarian tissue was identified. The surface of the cystic structure was smooth, bluish-grey and faintly glistening. There were no adhesions. In many areas, distended thin-walled subserosal vessels were present.

On cut surface, the cyst was multilocular, with individual compartments varying considerably in size and shape (Fig. 1). The locules were filled with turbid non-mucinous fluid. There were no papillary excrescenses. More solid areas of reddish-grey colour were encountered between the locules. The consistency varied from soft to firm and rubbery.

Both Fallopian tubes and the uterus were unremarkable. The left ovary was atrophic.

Histological findings.—Sections from various portions of the tumour were prepared and stained with hematoxylin-eosin and Masson's trichrome stain.

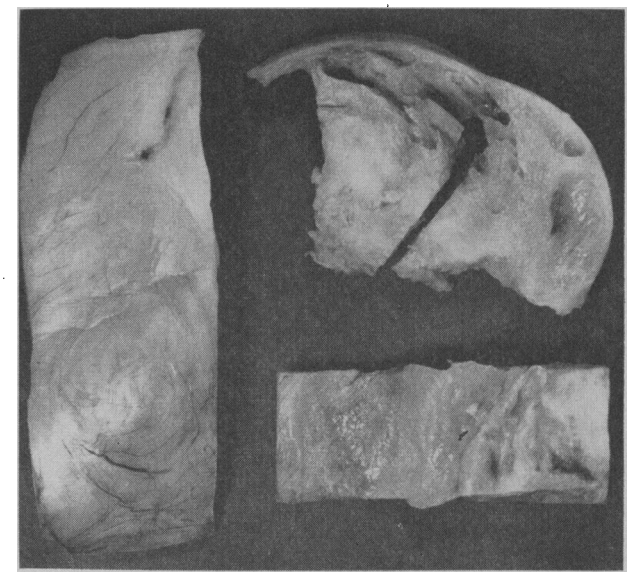

Fig. 1.—Close-up photograph of small portions of the tumour. The smooth external surface (left), a part of the wall of one of the locules from the interior of the tumour (lower right) and the cut surface with solid, porous and cystic portions (upper right) are shown.

In all of the sections, the bulk of the tissue was composed of typical smooth muscle cells with elongated blunt nuclei (Figs. 2 to 4). In the more solid

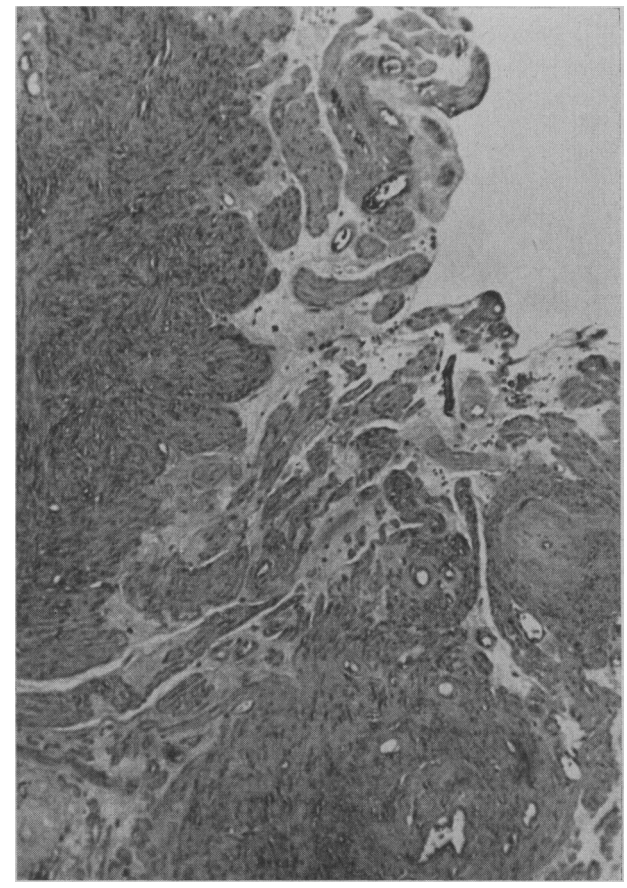

Fig. 2.—Microscopic picture of one of the more solid portions of the tumour.

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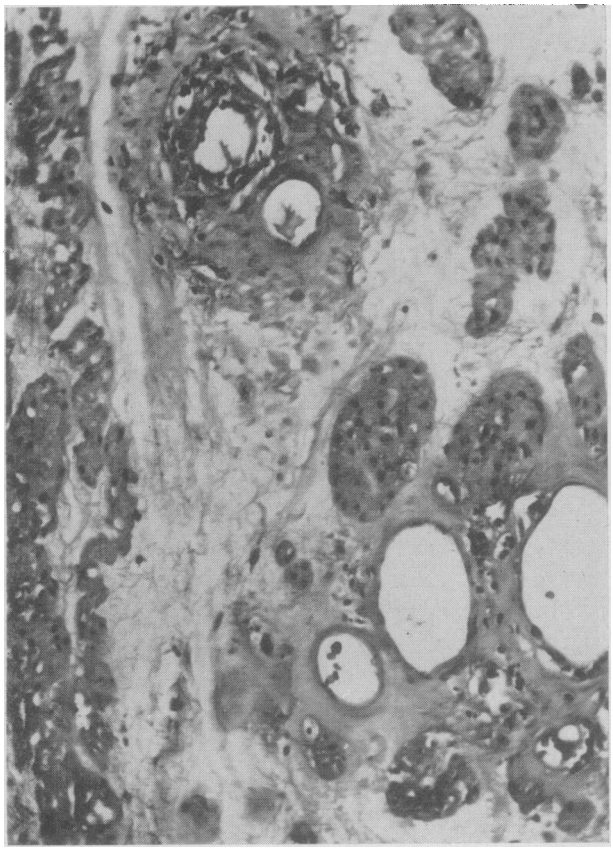

Fig. 3.—Islands of smooth muscle surrounded by loose-textured edematous tissue with but few formed elements. Small blood or lymph vessels are conspicuous in this field.

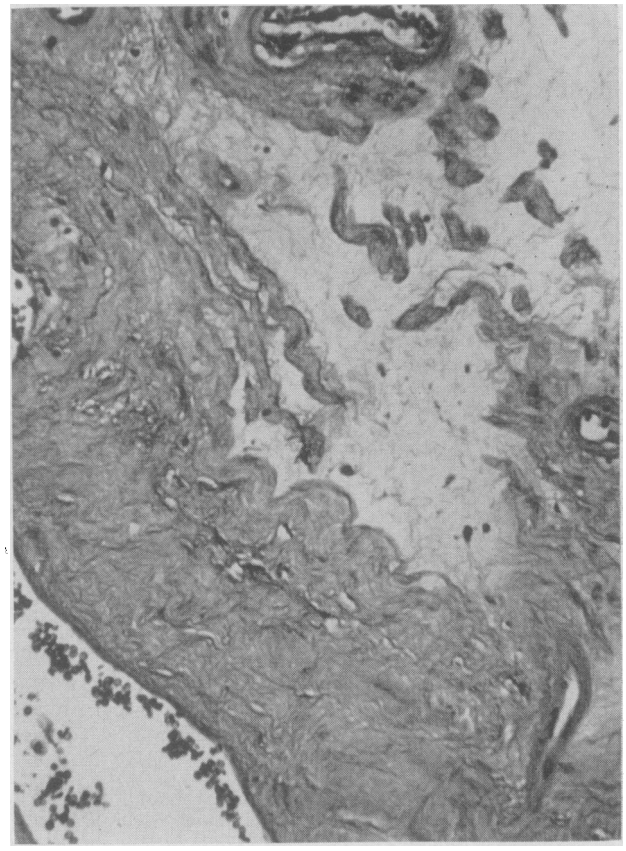

Fig. 4.—Wall of large blood vessel and a few small vessels without adventitial layer. Individual fibres of the muscle coat branch out to join those of the tumour between the vascular structures.

portions, they formed strands and interlacing bundles arranged in the whorled pattern characteristic of a leiomyoma. There was a gradual transition to fields extremely rich in intercellular edematous fluid, resulting in a wide separation of muscle bundles and individual fibres; the sarcoplasm of many of the latter was vacuolated. Dense focal aggregations of capillaries were noted. The walls of some of the larger vessels were rather thick and hyalinized. Very little connective tissue was present. Foci of hemorrhage and necrosis, with no cellular reaction, were encountered, particularly next to cystic spaces. Epithelial elements or normal ovarian tissue were not found. The endometrium showed the pattern of cystic atrophy.

DISCUSSION

In his comprehensive monograph on diseases of the ovary, Miller,[9] in 1937, commented on cases of ovarian leiomyoma reported by 29 different authors. He did not clearly distinguish between pure leiomyoma and fibromyoma, the latter tumour presenting a sizable amount of fibrous matrix along with the musculature. He did exclude, however, tumours in which the fibrous element predominated, i.e. myofibromas. Miller stated that pure leiomyomas of the ovary are "extreme rarities".

Kleitsman,[6] in 1950, found only 17 reported cases that he was willing to accept as pure ovarian leiomyomas, and added one of his own. Further cases were reported by Fino,[3] Herbut,[5] Massé, Dax and Carles,[8] and Moore.[10]

From a survey of the reported cases it becomes clear that the majority of ovarian leiomyomas are not accompanied by significant clinical symptoms. In pure leiomyomas, no menstrual irregularities are encountered; an exception to this observation was the case reported by Basilier (quoted from Kleitsman[6]). In fibromyomas, on the other hand, such irregularities are more frequently reported. Weight loss, ascites and increase of abdominal girth may complicate large and fairly rapidly growing tumours, but most leiomyomas of the ovaries are small, measuring only a few millimetres or centimetres in diameter. Apart from the present case, the largest acceptable neoplasm of this type and location was that of Henrotin and Herzog[4] (24 x 20 x 14.5 cm.); the tumour in Herbut's[5] patient had a maximum diameter of "approximately 25 cm.". Rarely, such tumours become twisted around their pedicle, with subsequent hemorrhage and necrosis. Most of the tumours reported were freely movable, but some became adherent to either intestine, uterus or, most frequently, omentum. All recorded ovarian leiomyomas were unilateral.

Moore,[10] in 1945, reported an ovarian leiomyoma complicating pregnancy and could find only two similar instances in the literature. In one of these cases, that of Olshausen, bilateral ovarian tumours are described, but microscopic proof is lacking. Another ovarian leiomyoma leading to delayed

delivery and postpartum hemorrhage, that of Münchmeyer, is mentioned by Miller.[9]

All tumours of this type are described as being rounded and more or less solid and firm, although they tend to be softer than fibromas. The ovary proper may become completely absorbed by the tumour, as in our case, or portions of it may persist. A reddish colour, rather than the greyish-white colour of fibromas, is considered characteristic. Frequently, whorl formation is grossly recognizable. Some tumours are rich in lymph and blood vessels. Seven of the 18 ovarian leiomyomas surveyed by Kleitsman[6] were accompanied by identical tumours of the myometrium.

Secondary alterations, such as hyalinization, hemorrhage, calcification, edematous imbibition and cyst formation, are probably present, to a minor degree, in the majority of all larger tumours of this kind. However, only one other case of an ovarian leiomyoma, that of Massé, Dax and Carles,[8] has been found in which cystic degeneration was extensive enough to dominate the gross appearance of the tumour. In both cases, it was only after histological sections were studied that the true nature of the tumour was recognized.

Certain points in the case reported by Massé, Dax and Carles[8] remain open to question. The authors do not state the exact origin of this neoplasm and concede that it may have developed in the broad ligament. Nothing is mentioned about the ovary proper. Very unusual for an allegedly benign tumour was the presence of a fistula between small bowel and one of the locules of the tumour, necessitating the removal of 40 cm. of bowel. The gross impression was that of a "malignant tumour of cystoepitheliomatous type". The 65-year-old patient died one year after the operation, with jaundice. Autopsy findings are not recorded.

Obviously, progressive degeneration and cyst formation, with accumulation of more and more fluid, rather than active growth of tumour cells, led to the rapid enlargement of the neoplasm reported in this paper. Leiomyosarcoma of the ovary, as another theoretical explanation for rapid growth, is exceedingly rare, only one case being known at this time.[9]

The histogenesis of ovarian leiomyoma is not fully understood. Three theories have been advanced in the past. The first relates the origin of the tumour to smooth muscle fibres in the medulla and hilus of the ovary.[2, 7]

According to Wallart,[12] two more or less separate muscle layers may be distinguished in the mesovarium, the one subserosal, the other between the vascular structures traversing the mesovarium. From the latter, the so-called intervascular layer of smooth musculature, muscle fibres extend into the hilus of the ovary and further on into the medulla but not into the cortex. In infants and children, and after the menopause, the number of smooth muscle fibres is less than in the fertile period of life, while there are marked hypertrophy and hyperplasia of ovarian and mesovarian musculature during pregnancy. Lesser degrees of hyperplasia and hypertrophy may be found in cases with coexisting uterine leiomyoma and, according to Martella (quoted from Kleitsman[6]), in chronic oophoritis.

A second hypothesis refers to smooth muscle fibres in vessel walls as the point of origin.[4]

More recently, Kleitsman,[6] referring to the older concept of Frankl and Robert Meyer, postulates the presence of "originally undifferentiated cell germs" in the ovarian stroma which, according to him, may become stimulated to differentiate into smooth muscle, with ultimate tumour formation. The stimulating event is considered to be a hormonal disturbance, such as overproduction of folliculin.

Another possibility is the development of ovarian leiomyomas from muscular structures of vestigial remnants of the Wolffian body, e.g., Kobelt's tubules. Finally, they may presumably arise from foci of endometriosis. The presence of muscle fibres in islands of endometriosis in the ovary is very rare but has been demonstrated occasionally; Miller[9] lists six such cases. In Kleitsman's[6] case and in another one mentioned by Miller (that of Chomolgorow) the contralateral ovary was the seat of what was termed "adenomyosis".

Most observers favour Doran's[2] and Krömer's[7] theory which postulates an origin from pre-existing smooth muscle fibres not related to blood vessels. It may be, however, that not all leiomyomas of the ovary originate in an identical manner. Henrotin's and Herzog's[4] hypothesis, that the tumour is from vascular musculature, still awaits refutation. Thus, in some of our sections, medium-sized and smaller blood vessels were encountered with rather thin muscle coats but completely lacking adventitial layers. From the deficient muscle coat, individual fibres frequently branched out to join those of the tumour, and it was impossible to decide which of the muscle fibres were part of the tumour and which belonged to the vessel wall (Fig. 4). Basso,[1] in 1905, published identical findings in his four cases of leiomyoma or fibromyoma.

In the discussion of the development of ovarian and uterine leiomyomas, hormonal influences are given a great deal of consideration. In keeping with this emphasis on the role of hormonal influence, which is supported by animal experiments, is the fact that the great majority of leiomyomas of these two organs grow only between puberty and the menopause. However, a number of cases remain in which this rule does not apply. More recently, for instance, Zeit[13] has reported on such myomas occuring during and after the menopause. Hormonal disturbances are not always elicited in these patients.

With two exceptions, all reported cases of ovarian leiomyoma were in women in the childbearing age; in a number of cases the age was not listed. Miller[9] gives 20 and 53 years as age limits;

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Kleitsman,[6] 22 and 52. The tumour was found in nulliparous women as well as in mothers of many children. The two exceptions in age are the cases reported by Godlewsky (quoted from Kleitsman[6]) and Massé, Dax and Carles;[8] the case report of the latter is of a 65-year-old woman.

Our case appears to be the third instance of an ovarian leiomyoma developing in a postmenopausal woman. Clinically, there were no symptoms pointing to a possible hormonal derangement in this patient, either before or in the months after the operation. Furthermore, the finding of an atrophic endometrium at the time of operation militates against the assumption of hyperestrogenism as a stimulus leading to the growth of this particular tumour.

SUMMARY

The case of a pure leiomyoma of the ovary is described which, apart from the extreme rarity of this tumour in this location, presented several unusual features: (1) the tumour is the largest reported in the literature; (2) it showed pronounced cystic change, thus imitating the clinical and gross pathological appearance of a multilocular cystadenoma; (3) the extensive cyst formation of this neoplasm was probably responsible for its rapid growth; and (4) it is the third known case of an ovarian leiomyoma manifesting itself in a postmenopausal woman.

A short review of the literature is presented, and pertinent features of the symptomatology, pathology and possible histogenesis of ovarian leiomyoma are discussed.

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VENTRICULAR FIBRILLATION IN A BURNED BOY

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THE OCCURRENCE of cardiac arrest (standstill and ventricular fibrillation) associated with the burned state appears to be more common than is generally realized.[1-5, 8, 9] Moreover, two episodes of cardiac arrest within one week in the same child have been reported from both Australia[4] and Sweden.[3] In each of these patients arrest occurred during the induction of an anesthetic between the 28th and 42nd day after the burn, and was in each instance preceded by the administration of several uneventful anesthetics.

This report concerns a burned child in whom two incidents of cardiac arrest occurred within seven days. It seemed advisable to record this case and to provoke further thought that may lead to a better understanding of the mechanisms which may be peculiar to cardiac arrest in the burned patient.

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A boy, aged 14 years (weight 93 lb., height 5′ 2″), was admitted to hospital with severe burns from the ignition of his gasoline-soaked clothes. Approximately 36% of the body surface was involved, about half of which was third degree and the remainder second degree. The burned area involved the whole of the left upper extremity, the left side of the trunk, the posterior aspect of both thighs and both lower legs except for the feet. The perineum was free.

He was treated with dextran, blood and other intravenous fluids for the first four days. Urine output was checked by catheter collection for five days. Subsequently he received blood and plasma as indicated by the degree of hemoconcentration, hemodilution and fall in serum protein level. Electrolyte studies had been carried out during the initial period. An effort was made at all times to enforce a maximum intake. The use of a stomach tube and forced feedings were not tolerated. Antibiotics were given according to bacterial cultures and sensitivity. The predominant organisms throughout were *Staphylococcus pyogenes* and *Proteus vulgaris*. Neomycin was used locally on the dressings on several occasions. Although all burned areas that were not epithelialized were infected and showed purulent exudate, no grafts were lost. Throughout his entire hospital stay and for some time after discharge his pulse rate was more than 120 per minute, and his temperature varied between 99° and 101° F. Electrocardiograms showed only sinus tachycardia.