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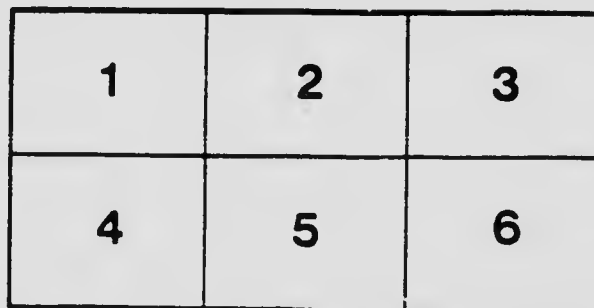
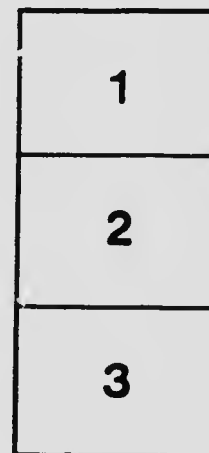
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An Extra-Abdominal Multilocular Ovarian Cyst

THOMAS S. CULLEN, M.D.
BALTIMORE

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AN EXTRA-ABDOMINAL MULTILOCLAR OVARIAN CYST*

THOMAS S. CULLEN, M.D.

BALTIMORE

Several months ago, Dr. Frank R. Smith asked me to see a woman who had a kidney-shaped tumor slightly below and to the right of the umbilicus. The patient had noticed a small lump in this situation several years before, which for a long while had remained quiescent but during the last year had gradually increased in size. At operation it was found to be a partly solid, partly cystic tumor of the ovary lying external to the abdominal muscles, the tumor and its surrounding sac being covered over with a small amount of adipose tissue and the skin. The pedicle of the tumor passed through a hernial ring to the outer side of the right rectus and obliquely across the lower abdominal cavity to what corresponded to the normal insertion of the right utero-ovarian ligament.

I have been unable to find any reference to a similar case in the literature.

History.—Mrs. M. W., aged 56, was a short, well developed woman, and, apart from a tumor mass in the lower abdomen, was in excellent health. She had had eleven children. Her periods had ceased at 50. She had felt some pain in the right ovarian region for fifteen years, and for about ten years she had noticed a little tumor situated in the right lateral abdominal wall slightly above a line drawn between the umbilicus and the anterior superior spine. This, from her description, seemed to have been about the size of an ovary. During the last ten months this small lump had increased in size until it formed a lobulated mass, elongate in form, about 10 by 8 cm. It seemed to be but a short distance beneath the skin and could be lifted up to some extent in the hand, but its absolute relationship could not be determined on account of the presence of a considerable amount of adipose tissue.

* From the Gynecological Department of the Johns Hopkins Hospital.

Operation.—Nov. 1, 1910, on making a pelvic examination, under anesthesia I found that the uterus was normal and that there was no thickening laterally. Not being sure of the exact condition, I made a median incision. The uterus was normal, the left tube and ovary presented the usual appearance. The right tube showed no change, but the right utero-ovarian ligament was markedly drawn out into a band about 1 cm. broad. This led to a hernial opening with smooth margins in the right lateral abdominal wall below and to the right of the umbilicus, but at least 12 cm. from the inguinal region (Fig. 1). After obtaining good exposure I found that this flattened band of the utero-ovarian ligament passed directly into a hernial opening about 2.5 cm. in diameter, and into this opening a finger could readily be introduced. The intra-abdominal portion of the pedicle was clamped off and sutured. An incision was then made over the prominent part of the abdominal tumor, which proved to be extra-abdominal. The more prominent part of this tumor lay directly beneath the skin in the adipose tissue, and was very easily freed by blunt dissection to the point where the hernial ring entered the abdomen. I then cut the peritoneum around the hernial ring and delivered the tumor, with its peritoneal covering intact. The space where the tumor had existed having been obliterated and the inner incision having been sutured, the outer wound was also closed. The ovarian tumor was multilocular.

There had evidently been a hernial protrusion through the right lateral abdominal wall, into which the ovary had dropped and remained for several years. During the last year it had increased in size and given rise to a multilocular ovarian cyst. Naturally with the increase in size, the escape of the ovary from the sac was impossible.

Macroscopic Examination of Hardened Specimen (Path. No. 15,723).—The hernial opening was about 2.5 cm. in diameter. Its margins consisted of peritoneum, outside of which was a zone of adipose tissue. The tumor itself was kidney-shaped (Fig. 2) 12 cm. long, 7 cm. broad, and 6 cm. in thickness. It was covered everywhere with peritoneum, which could be readily separated from it. Here and there attached to the outer surface of the peritoneum were tags of adipose tissue. The tumor itself was in large measure solid, resembling a fibroma. It presented a lobulated appearance. Here and there between nodules it showed cystic spaces, oblong, irregular, or round, varying from 2 mm. to 2 cm. in diameter. The majority of these were transparent and contained clear fluid. Some of them were slightly blood-tinged. So much could be made out through a window, which was cut in the peritoneum. On peeping in through the hernial ring, were seen cysts varying from 2.5 cm. to 3 cm. in diameter and apparently filled with clear fluid. After the drawing had been made the tumor was cut in two. The appearance on section is well shown in the drawing in the right upper corner of Figure 2.

Histologic Examination.—The solid portion of the tumor consisted in large part of fibrous tissue containing triangular or spindle-shaped nuclei. In some places the nuclei were abundant, in others scanty in number. The tissue showed a considerable degree of hyaline degeneration. At one or two points characteristic ovarian stroma was still in evidence. No Graafian follicles could be found, but after an examination of numerous sections a typical corpus fibrosum was noted. In some sections a few bundles of non-striped muscle were visible. The stroma had a meager blood-supply except in a few areas, where there were groups of rather large veins.

Scattered sparingly through the stroma were small circular or irregular glands occurring singly or in groups of two or three. They were found to be lined with cylindrical epithelium and were similar to those so frequently noted in the bilium of the ovary. Some of the very small cystic spaces, noted macroscopically, were lined with cylindrical ciliated epithelium and had an underlying stroma that stained rather deeply and that consisted of cells with oval vesicular nuclei. This stroma stood out in sharp contrast to the surrounding fibrous tissue. Such cysts frequently contained a little fairly fresh blood. These cysts reminded one very much of the cystic spaces so frequently noted in an adenomyoma, but I believe that they represented only the earlier stages of the larger cysts.

The large cysts were lined with one layer of epithelium which might be cylindrical, cuboidal or almost flat. Projecting into some of the cysts were papillary folds. These occasionally occurred as delicate, irregular, finger-like projections, but in the main as blunt, single or branching outgrowths. All of them were covered over with one layer of epithelium. The stroma of the papillary masses had in many places undergone almost complete hyaline degeneration, and in a few liquefaction of this hyaline material had taken place. Even in some of the larger cysts a moderate amount of fresh blood was present. The stroma cells beneath the cyst epithelium had in some places become swollen and spherical and were filled with yellow or brown pigment indicating the absorption of blood at some previous time.

On the surface of the tumor were a moderate number of vascular adhesions, and on the under and protected side of these the peritoneal cells had become cuboidal as is common on the under side of tubal or ovarian adhesions.

From the above description it will be seen that the dense matrix of the tumor consisted essentially of fibrous tissue and that scattered throughout this were multiple cysts, in large measure similar in character, some of which had small papillary masses projecting into them. Had the tumor developed in the abdominal cavity, I believe that in all probability it would have been a multilocular cystoma, but as it lay between the abdominal muscles and skin a rapid cystic growth was much

more difficult, and the fibrous tissue was thus allowed to keep pace with the cystic formation.

There was no sign of malignancy.

THE PRESENCE OF OVARIES IN HERNIAL SACS

The occurrence of ovaries and ovarian tumors in hernial sacs is of particular interest in this connection. I shall, therefore, briefly discuss the more important contributions made in this field.



FIG. 1.—OVARIAN PEDICLE PASSING FROM UTERUS, OUT THROUGH A HERNIAL RING IN THE ABDOMINAL WALL. A schematic representation of the pelvic structures as found at operation. The uterus and left appendages were normal. The right tube was unaltered, but passing from the uterus where the right ovary should have been was a band 1 cm. broad. This ran upward and outward and passed out through an abdominal ring to the outer side of the right rectus. At the ring a finger could be passed completely around this pedicle. It was nowhere adherent to the ring. A glance at the umbilicus will indicate the exact location of the ring. On looking through the ring a small portion of the glistening tumor could be readily seen. The dotted line indicates the relative size of the tumor.

The ovary has a rather free excursion, and it is not to be wondered at that where an inguinal or femoral hernia exists the ovary and often the tube form part of the contents of the hernial sac. Puech¹ in 1879 contributed a most interesting article on hernia of the ovary. In addition to his own cases, he refers to those of Pott, Lassus, Deneux, Guersant, Parker, Brun, Warren, Meadows, Cusco, Coote, Englisch, Weinlechner, MacCluer, Sonnenburg, Werth and Rheinstaedter. In brief his paper says that the ovary may pass out at any of the natural abdominal openings, and that, when it does so, it is most frequently by way of the femoral or inguinal ring. The inguinal hernia, which is the more frequent, may be congenital or accidental. The congenital hernia is produced by a process analogous to the descent of the testicle. In twenty-eight cases of hernia containing the ovary, fourteen were incontestably congenital, seventeen doubtful, and seventeen accidental. The congenital hernias were unilateral in twenty-seven cases, bilateral in twenty-seven cases. In the unilateral cases the left side was more frequently involved. In cases of double hernia the presence of the ovaries in the hernial sac was sometimes associated with defective development of the generative organs. Puech had records of sixteen cases in which the ovary was contained in a femoral hernia. In eleven of these cases the hernia was on the right side and in four on the left.

The uterus or one of its cornua was found ten times in the sac in cases of inguinal hernias, three times among the femoral hernias.

Dr. Joseph C. Bloodgood tells me that he has had two cases of femoral hernia with the ovary lying in the hernial sac.

Several years ago I operated on a child in Cambridge for an inguinal hernia. The sac contained the right tube and ovary. They were so intimately blended with the sac that it was impossible to save them, and they were removed preparatory to doing a radical operation.²

1. Puech, A.: Nouvelles recherches sur les hernies de l'ovaire, *Ann. de gynec.*, Paris, 1879, xi, 401.

2. This case was reported and illustrated in an article published by Dr. Frank T. Andrews (*Hernia of the Ovary and Tube*, *THE JOURNAL A. M. A.*, Nov. 24, 1906, p. 1707). Dr. Andrews' article gives an excellent résumé of hernia of the ovary and tube, and likewise contains a long tabulation of cases heretofore observed.

My experience with ovaries in hernial rings associated with lack of development of the generative organs has been limited to one case.³

A postoperative hernia occasionally contains an ovary. I reported a case of this character several years ago.⁴ The patient had had an appendix abscess, which it had been necessary to drain liberally. The resultant hernia in the scar was very tender and in dissecting it out I found that the sensitiveness was due to the presence of the right ovary contained in and adherent to the hernial sac.

OVARIAN TUMORS DEVELOPING IN THE INGUINAL CANAL

Since one or both ovaries may occasionally be found in the inguinal canal, we should not be surprised to find now and then a case in which such an ovary undergoes tumor development. Rheinstaedter⁵ in 1878 reported the case of a woman, 68 years of age, who had an elongate oval tumor in the neighborhood of the clitoris. In addition, there was a right-sided globular mass, which was found to be a right inguinal hernia. The oval tumor was larger than a child's head, elastic and apparently fluctuant. The overlying skin was normal. A probable diagnosis of ovarian tumor in a left inguinal hernia was made. Six months later the left hernial sac was opened. The tumor was easily raised and its broad pedicle apparently containing the tube and round ligament was tied off, some adherent loops of small bowel were loosened and pushed up into the abdomen, and the wound closed. The tumor weighed 750 gm. Careful examination of the right hernia revealed the presence of the ovary in the sac.

Marchand, who made the microscopic examination, diagnosed the tumor as an angiosarcoma.

Rheinstaedter, in reviewing the literature on the subject, was able to find only one case in which an ovarian tumor in an inguinal hernia reached the size of a child's head. This was the case observed by Cazati and quoted by Beigel. The hernial sac contained a portion of an ovarian cyst, the remainder of which lay in the pelvis.

3. Cullen, Thomas S.: A Right Pelvic Kidney; Absence of the Left Kidney; Absence of the Uterus, Both Ovaries in the Inguinal Canals, *Surg. Gynec. and Obst.*, July, 1910, p. 73.

4. Cullen, Thomas S.: The Right Ovary in the Abdominal Scar Following an Operation for Appendix Abscess, May, 1906, *Bull. Johns Hopkins Hosp.*, xvii, No. 182, p. 152.

5. Rheinstaedter: Kindskopfgrosses Angiosarkoma Ovarii in einem Leistenbruch, *Centralbl. f. Gynäk.*, 1878, II, 343.



Fig. 2.—EXTRA-ABDOMINAL MULTILOCULAR FIBROCYSTOMA OF THE OVARY. The dotted lines indicate the pelvic structures and the right ovarian pedicle passing upward and outward until it emerges from the hernial ring, a short distance below and to the outer side of the umbilicus. After the abdominal relations had been determined an incision was made directly over the tumor and it and its peritoneal covering were removed intact. A thin layer of fascia and the peritoneum formed the sac. The tumor was roughly kidney-shaped, lobulated and consisted partly of a solid tissue, partly of cysts. Deep clefts subdivided the tumor into several portions. The tumor in the main was free from adhesions, but in a few places there were points of union between the tumor and the peritoneal covering. The drawing in the right upper corner shows the tumor on section. The fibrous tissue is abundant, but at this level the cysts predominate. The clefts are seen to have extended through the tumor. It was possible to lift the various segments out without disturbing the remaining ones. Figure 3 gives the real form of the tumor when liberated from pressure.

Of solid ovarian tumors contained in hernias he mentions only a case of Guersant reported by Englisch.

Fargas,⁶ in 1890, described the case of a woman, aged 38, who had a tumor the size of a man's head occupying the right inguinal and labial region. On the left side was an inguinal hernia containing an ovary, which was easily reducible. The patient said that until eight years previous the right side had presented the same appearance as the left, and Fargas, therefore, diagnosed the tumor as a right-sided ovarian cyst. Six years prior to observation the right hernia had become irreducible, and after that had gradually increased in size.

The hernial sac was opened and the cyst lay free from adhesions. Its pedicle was tied off as in an ordinary ovariectomy and the tumor removed. The patient made a good recovery. The cyst was reticulated (multilocular). Fargas confessed that it might have been confounded with a cyst of the round ligament, but its topography rendered its source certain. The woman had had normal menstruation and had borne two children during the existence of this tumor.

Seymour⁷ in 1897 reported the case of a woman aged 48. Two years before coming under observation a small tumor had appeared in the right groin. It was somewhat sensitive, hard and irreducible. An inguinal hernia with possibly an incarcerated omentum without strangulation was thought of. At operation the tumor was found attached to the uterus by a pedicle, 4 inches long and $\frac{3}{4}$ inch in diameter. This passed out through the inguinal ring to the under surface of the upper end of the tumor in the inguinal canal. The pedicle was tied off and the sac closed. The tumor was ovoid in shape, 4 inches in length and 8 inches in circumference. The microscopic report was spindle-celled sarcoma.⁸

6. Fargas: Note sur un cas de hernie inguino-labiale de l'ovaire droit avec gros kyste réticulaire du même organe, Arch. de tocol., Paris, 1890, xvii, 767.

7. Seymour: Hernia of a Sarcomatous Ovary, Tr. Med. Soc. State of N. Y., 1897, p. 370.

8. If the fibrous character of a solid ovarian tumor is definite, one is safe in making a diagnosis of fibroma, and if the growth consists of spindle cells and has many mitotic figures and atypical cell changes, one can with justice make a diagnosis of sarcoma. There are, however, quite a number of solid tumors of the ovary of the spindle-celled type, in which it is impossible to say from the microscopic examination whether the growth is a sarcoma or a fibroma. In such cases the diagnosis can never be definitely determined unless the patient subsequently develops metastases.

AN OVARIAN CYST PROTRUDING INTO THE INGUINAL
CANAL

Baldy^o in 1898 reported an interesting case of this character. Two years before coming under Baldy's observation the patient had noticed a small pedunculated tumor in the right inguinal region. This had continued to grow until it was as large as an egg and had then been removed. A year later a larger and similar growth had been present and had also been removed.



Fig. 3.—AN EXTRA-ABDOMINAL MULTILOCCULAR FIBROCYSTOMA. A schematic representation of the manner in which the tumor tended to develop when relieved from its surrounding pressure. It in reality consisted of four lobes similar in character and joined together by broad or narrow pedicles.

When Baldy saw the patient the growth was as large as a fist. A pelvic examination revealed a mass filling the right side of the pelvis.

At operation, an incision was made directly over the growth and the tumor freed down to a thick, broad.

^o Baldy, J. M.: Ovarian Cyst Protruding Through the Inguinal Canal, *Am. Jour. Obst.*, 1898, xxxviii, 827.

short pedicle which passed into and through the inguinal canal. The attachments of the pedicle in the canal were freed and the finger forced into the abdomen when it was found that the tumor was a continuation of a large intra-abdominal mass.

When the abdomen was opened in the median line, the pedicle of the inguinal growth was found springing from a large intraligamentary tumor similar in character. The left uterine appendages were normal. A supravaginal hysterectomy was performed and the hernial ring closed. The patient made an uninterrupted recovery.

AN OVARIAN CYST LYING PARTIALLY IN A FEMORAL HERNIA

This condition is exceptionally rare. Dellhaes¹⁰ in 1885 cited the case of a woman, aged 55, who entered the hospital in 1879 on account of ascites. For four years she had complained of discomfort in the lower abdomen, and there had been an increase in size in the right side. Thereafter she had had a crural hernia on the right side and for the last nine months a similar hernia on the left. The right was reducible, the left irreducible.

After the patient had been built up to some extent and after the ascitic fluid had been removed several times by puncture, a right-sided ovarian tumor, partly solid, partly cystic, was removed.

The left ovary was cystic and enlarged. It was partly situated in the pelvis and had to be separated from a solid cord which passed into the femoral canal. The major portion of the large cystic left ovary lay in the fold of the groin and fourteen days later was removed through a herniotomy incision. This portion was as large as a child's head. The patient made a complete recovery.

A SMALL PORTION OF AN OVARIAN CYST INCARCERATED IN THE ABDOMINAL WALL JUST BELOW THE UMBILICUS

Hopkins¹¹ in 1894 reported an interesting emergency case. The patient, aged 57, was admitted for a supposed strangulated umbilical hernia. Protruding just

10. Dellhaes: Eine Hernia crurails cystovarii sinistra. *Ztschr. f. Geburtsh. u. Gynäk.*, 1885, xi, 389.

11. Hopkins, George G.: An Ovarian Cyst Simulating a Strangulated Umbilical Hernia. *Boston Med. and Surg. Jour.*, July 26, 1894, p. 84.

below the umbilical opening was an almost black tumor the size of a walnut. Still thinking that it was a strangulated hernia, the operator cut the constricting band and the tumor at once disappeared. The abdominal opening was enlarged and serous and gelatinous material began to pour out. On further exploration a multilocular ovarian cyst, weighing about 25 pounds, was found. This was removed and the patient promptly recovered.

A small portion of this cyst had become strangulated in a small hernial defect just below the umbilicus.

DEFECTS IN THE ABDOMINAL WALL

A study of the weak spots in the abdominal wall is of interest. Levadoux¹² made a most exhaustive examination of the inner appearance of the umbilicus, of the final disposition of the umbilical arteries, the umbilical vein and the urachus, and described in detail how these sometimes blended in such a manner as to form a fibrous sheath that completely covered over the inner umbilical opening. In the course of his studies he also noted weak spots in the fascia just beneath the peritoneum. These openings were usually small and were oval, round or irregular in shape.

On careful scrutiny it has at times been noted that hernias, supposed to be umbilical, were in reality peri-umbilical and that the umbilicus itself was intact. These defects in the fascia are undoubtedly the cause of such hernias. In July, 1910, I saw, in consultation with Dr. A. H. A. Mayer, a boy, aged 17, who had a small hernial protrusion 4 cm. above and to the left of the umbilicus. This hernial protrusion projected 1 cm. through the fascia and was lobulated, forming a mass 3 cm. in diameter. The patient was of spare build. On cutting down on the hernial sac I found a small defect in the abdominal wall with a small portion of the omentum projecting through it. The omentum was readily returned and the opening easily obliterated with a few sutures.

Dr. Bloodgood tells me that he observed a case of hernia of the abdominal wall at the splanchnic line, that was between the rectus muscle and the transverse forming the

12. Levadoux, Michel-Joseph: Variétés de l'ombilic et de ses annexes. Thèse Fac. de Méd. et d'Pharm. de Toulouse, 1907, No. 711.

lateral abdominal wall. The sac contained non-adherent loops of small bowel. The condition was readily cured. In our case the opening was also at, or near, the semilunar line, and instead of small bowel passing into it the ovary had for some reason occupied the space and later had gone on to tumor development. It is just possible that this weakness in the wall had become particularly accentuated during a pregnancy and that the ovary during its ascent with the pregnant uterus had dropped into the cavity.

In conclusion, I wish to thank Dr. Fred Ray for his kindness in ferreting out the references to the articles read by me in the preparation of this paper, and I am especially indebted to Mr. Max Brödel and the Foundation of Art in Medicine of the Johns Hopkins Medical School for the excellent illustrations.

