

Adenomyoma of the Rectogenital Space associated with Tarry Cysts arising in Islands of Adenomyomatous Tissue in the Ovary.¹

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At the May meeting of the North of England Gynæcological Society, Dr. Donald¹ read a paper on five cases of adenomyoma of the uterus, associated, in three instances, with tarry cysts of the ovary, in which he had operated within a period of three months. For a long time he had been struck with the frequent association of these two conditions, and concluded, on clinical grounds, that the tarry cysts probably originated in adenomyomatous tissue in the ovary, the dark contents being retained menstrual blood. Although a large number of these cysts had, from time to time, been examined microscopically at St. Mary's Hospital, no confirmation of this hypothesis could be obtained.

Lately, however, proof of the truth of his conclusions has come to hand in papers by Cullen² and Sampson,³ both of whom describe similar cysts as originating in islands of endometrial tissue in the ovaries.

To us, who have long known Dr. Donald's opinion on this subject, based as it was entirely on clinical observation, it is of particular interest that one of the first two cases in this country to confirm his hypothesis should come from his own school.

During the three months in which Dr. Donald's cases occurred, one of us operated on three cases of adenomyoma of the rectogenital space, two of which were associated with tarry ovarian cysts, and a few days after the reading of the paper operated on a fourth case which we now describe.

Mrs. G. M. (38 years of age, married, no children) was sent to one of us because of severe dysmenorrhœa of one year's duration. Examination revealed a firmly fixed irregular tumour, apparently arising from the uterus, filling the pelvis and extending up into the abdominal cavity to four fingers' breadth above the pubes.

1. Read at the British Congress of Gynæcology in Liverpool, July 1, 1922.



FIG. 1.



FIG. 2.



FIG. 3.

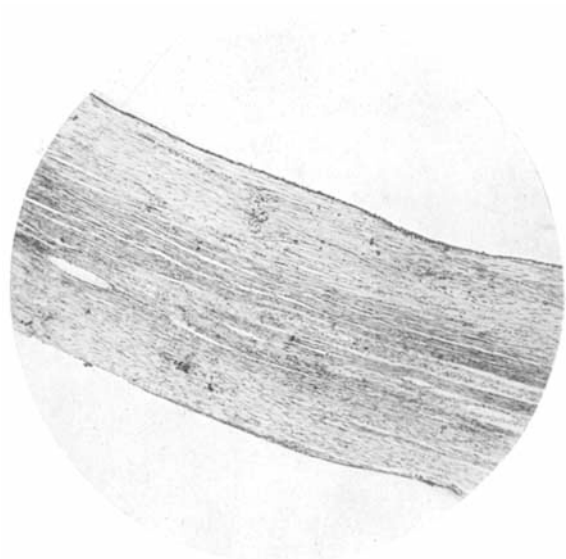


FIG. 4.

For six weeks prior to examination the right leg had been swollen, and there had been slight pyrexia at the last two periods. A diagnosis of fibromyomata of the uterus with inflamed and adherent appendages and parametritis was made. The patient was operated upon on May 6th, 1922.

When the abdomen was opened the tumour was found to consist of a mass of fibroids in the uterine wall, the right ovary, which was placed above the pelvic brim, was adherent to the bowel on its under surface, and the left ovary was buried in adhesions obliterating the pouch of Douglas.

As Cullen remarks, adenomyoma of the recto-vaginal septum frequently presents greater difficulty in removal than does a carcinoma of the cervix. In this instance the difficulties were considerably increased by the adherent condition of the ovaries, and removal was only accomplished after a long and arduous dissection, during which the rectum was found to be firmly attached to the back of the cervix by a dense bridge of tissue. A low supra-vaginal hysterectomy with removal of both appendages was performed. In spite of the severity of the operation the patient made an uninterrupted recovery.

On examination the uterus was found to contain a large number of fibroids, varying in size, and forming an irregular, globular mass the size of a swede turnip. The lower half of the globe was covered by firm adhesions and there was a nodular thickening at the back of the uterus about the level of the internal os, which on microscopic examination proved to be typical adenomyoma invading the uterus. (Fig. 1.)

The left ovary, which was the size of a tangerine orange, showed well organized adhesions over the whole surface. On section the ovarian substance was found to be almost entirely replaced by a series of thick-walled cysts, containing dark chocolate-coloured fluid of a tarry consistence. The left tube was separated from the ovary and, on microscopic examination, appeared healthy.

The right ovary, which was slightly smaller, and adherent only on its under surface, was seen, on section, to consist of two main cysts with contents similar to those in the left ovary, and a solid portion containing several small cysts of a like nature. The tube on this side was also free from adhesions and contained no adenomatous tissue.

Two blocks for section were taken from the right ovary. Each included, along with the solid portion, the lining of one of the large cysts, and was at least one inch from the ovarian ligament and about half an inch from the hilum.

The first of these (Fig. 2) showed gland spaces lined with a single layer of cubical epithelium, supported by an embryonic

stroma, indistinguishable microscopically from that of the endometrium, and lying on a layer of unstriped muscle.

The second (Fig. 3) showed a mass of endometrial stroma supporting the epithelial lining of the large cyst. On tracing this membrane round the cyst wall we found the stroma gradually to thin out and ultimately to disappear, leaving an unsupported epithelium which in turn became flattened out until it had lost all its distinctive characteristics as is seen in Fig. 4.

It is probable that the endometrium in this section had been gradually thinned out by pressure from the monthly addition of menstrual blood to the cyst cavity, and that if the condition had been allowed to proceed a little longer, the remnant of endometrium here shown would have entirely disappeared, leaving only a flattened endothelium-like lining.

This, we think, explains the frequent failure in the past to demonstrate Dr. Donald's conclusions as to the origin of these cysts.

Sections from the left ovary, which was in a much more advanced stage of the condition, support this contention, in that a very extensive search had to be made before any adenomyomatous elements could be found. Sections were taken from all parts of this ovary, but in only one area, namely, that near the insertion of the ovarian ligament, were endometrial elements found (Fig. 5), and then only as the lining of one of the smaller cysts.

Sections at the hilum of the left ovary (Fig. 6) are of interest in showing what would appear to be an exceptionally large number of Wolffian tubules.

Cullen mentions nine sites in which adenomyomata are found and a tenth, the ovary, in which uterine mucosa had been detected. In our specimen, however, in addition to glands and stroma, there are definite layers of unstriped muscle, and thus it is brought into line with those tumours, found in other situations, described as adenomyomata.

In all previous situations in which adenomyomata have been found, unstriped muscle was a normal constituent. In the ovary, however, with the exception of the point of insertion of the ovarian ligament, and possibly at the hilum, there is a complete absence of such tissue. Thus it is clear that in this specimen the muscular elements are, equally with the endometrial, a new formation. So far as we can discover this condition has not yet been described as occurring in the ovary.

Sampson, to whom must be given the honour of first describing the true origin of these cysts, reports 23 specimens with tarry cysts in the ovaries. He considers the dark contents to be retained menstrual blood produced in the endometrial glands in the ovaries,

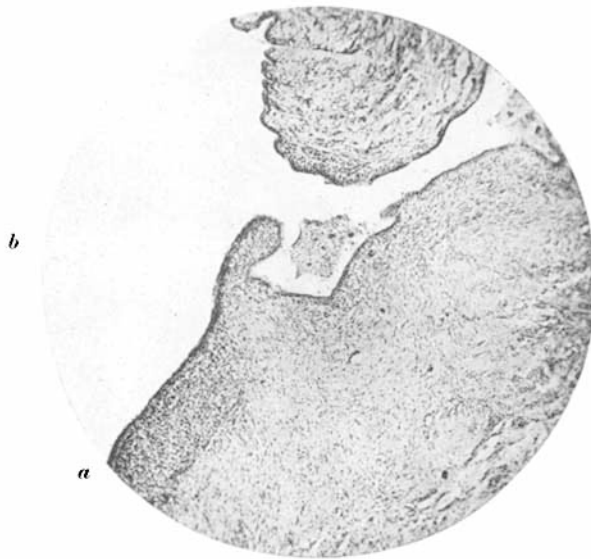


FIG. 5.

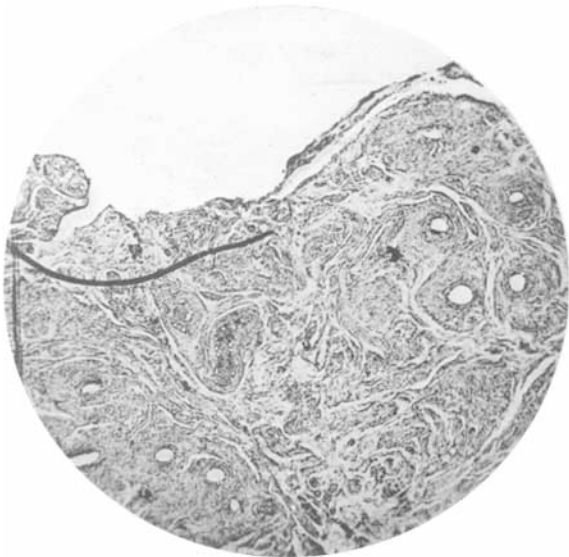


FIG. 6.



FIG. 7.

and the dense adhesions by which these ovaries are as a rule bound down, to be due to the irritation of this material which has burst through the cyst wall. This would account for the onset of dysmenorrhœa in our patient at the age of 37, though there is no account of previous pregnancy or inflammation.

We do not wish to discuss here the vexed question of the origin of these tumours, on which subject we feel much work remains to be done. The presence of muscle in this situation, however, clearly makes some of the hypotheses, so far advanced, untenable.

Adenomyoma is possibly not the best name for these tumours, but it has long been used since they were first described, and when we are dealing with a tumour consisting of endometrium and unstriped muscle we think it better to retain it, as it describes the constitution of the tumour sufficiently accurately, and so long as the ætiology is uncertain we do not see any advantage in further confusing the issue by adding to the terminology.

Like Sampson we believe these tumours will prove to be of common occurrence; the reason for their having been so long overlooked is, we think, explained by the hypothesis that the gradually increasing pressure in the cysts destroys the stroma and flattens out the epithelium, thus making microscopic diagnosis difficult and in advanced cases impossible.

Since preparing this paper we have operated upon two other patients with adenomyoma of the uterus associated with tarry cysts of the ovaries. In one of these no endometrial tissue could be demonstrated in the ovaries, probably because the pressure in the cysts had flattened it out; in the other case definite areas of endometrial tissue were seen in one of the ovaries (Fig. 7).

REFERENCES.

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2. Cullen, Thos. S. "The distribution of adenomyomas containing uterine mucosa." *Archives of Surgery*, i, 215—283.
3. Sampson, J. A. "Perforating hæmorrhage (chocolate) cysts of the ovary." *Abstract, Amer. Journ. of Obstet. and Gynecol.*, ii, 526.

Fig. 1. Section from back of uterus at level of internal os, showing typical adenomyoma invading uterine wall.

Fig. 2. Section from right ovary showing endometrial tissue supported by layer of unstriped muscle.

Fig. 3. Section of lining of large cyst in right ovary. An epithelial layer with supporting endometrial stroma.

Fig. 4. Section of lining of same cyst, showing disappearance of stroma and flattening of epithelial layer.

Fig. 5. Section of left ovary showing at *a* and *b* appearances similar to those seen in the right ovary of Fig. 3.

Fig. 6. Section from hilum of left ovary.

Fig. 7. Section from a second case showing adenomatous tissue in the ovary.