

On Adenomyoma of the Uterus,

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WE are unable to find any detailed or systematic account of the subject of adenomyoma of the uterus in British gynaecological literature. In consideration of this fact, and having recently had the opportunity of investigating a specimen typically characteristic of this condition we venture to place our case on record:—

Mary P——, a married woman, aged 49, was admitted on January 22nd, 1903, into Chelsea Hospital for Women under the care of Mr. Bland-Sutton, whom we wish to take this opportunity of thanking for his kind permission to publish this case. Until she was seven years old the patient was considered to be a delicate child. Menstruation commenced at the age of 13, and has always been regular,—with the exception of the physiological intermissions incidental to child-bearing—until between two and three years ago. At, and for some time subsequent to, the onset of menstruation there was a good deal of pain at the periods, but of late years there has been very little. The periods last a week, and there is no great loss. For between two and three years before coming under observation there had been gradually progressive menorrhagia, the loss becoming very profuse, and latterly containing large clots. For five weeks before operation there was profuse metrorrhagia. She had had four children, two miscarriages, and two “false conceptions,” the last pregnancy being six years ago. There has been no vaginal discharge between the periods, except that prior to 12 months ago she had a slight white discharge. There has never been any trouble with the functions of the bladder or bowels. Patient has become more and more anæmic with increasing metrorrhagia. On abdominal examination no pathological condition was determined. On vaginal examination the uterus was found to be enlarged, firm and freely mobile, as if occupied by a fibroid tumour. There was some “erosion” of the cervix and some sanguineous vaginal discharge. The diagnosis of fibromyoma of the uterus was made, and abdominal hysterectomy decided upon. On January 26th the abdomen was opened by a sub-umbilical median cœliotomy and the pelvis was explored. The uterus was found to be enlarged and to contain what appeared to be a fibromyoma about the size of a very small cocoonut. The uterus along with both appendages was removed, and only a very thin shaving of cervix was left behind. The two thin lips of cervix left behind were approximated with buried mattress silk sutures, and the peritoneum

was stitched up in the usual manner. The abdominal wound was sutured in three layers and no drainage was employed. Recovery was rapid and uneventful. The patient was discharged from the hospital cured on February 14th.

Pathological Report. The specimen consists of the enlarged uterus with one appendage attached, the other appendage having been removed at the operation. The measurements of the uterus are as follows:—Length, 5 inches; breadth, $3\frac{1}{4}$ inches; antero-posterior diameter, $2\frac{3}{4}$ inches. In shape it closely resembles a normal but enlarged uterus. Externally it is covered by peritoneum to a normal extent, the peritoneal covering being everywhere smooth and shining. Before section of the tumour it presented the appearances of, and was considered to be, an ordinary interstitial fibromyoma.

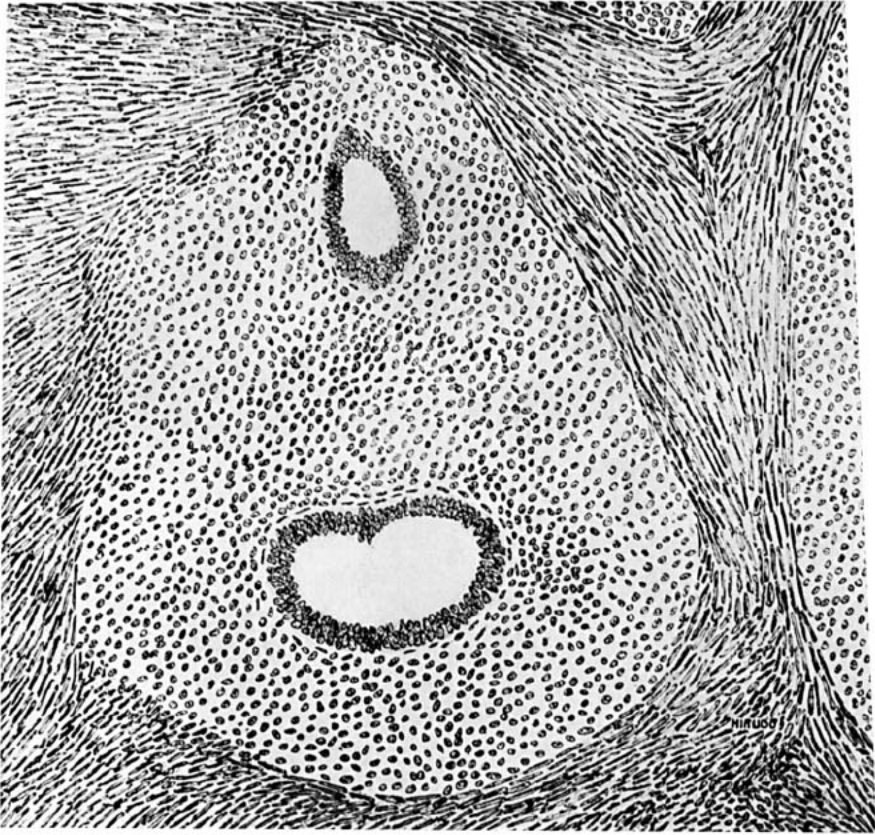
On making a section through the anterior wall of the uterus, however, a striking difference was observed. The tissues were hard to cut, and no trace of a capsule to the growth could be made out. The uterine wall was $1\frac{1}{4}$ inches in thickness, and could be readily differentiated into three layers, from within outwards, firstly, a thin layer of spongy, rather ragged endometrium; secondly, a mass of spongy-looking, coarsely striated tissue with striæ running in all directions, and not presenting the characteristic whorled appearance so typical of fibromyoma; and, thirdly, a layer of apparently normal uterine muscle about one-third of an inch in thickness. This appearance was present in the fundus and both anterior and posterior uterine walls as far down as a little above the os internum where the growth gradually faded away into the normal uterine tissues. Just beneath the peritoneum, where the growth and normal uterine tissues meet, was a small encapsuled nodule $\frac{1}{2}$ in. \times $\frac{1}{4}$ in. in diameter. It was pearly-white in colour, composed of concentrically arranged fibres, and presented, in fact, the macroscopic appearance of an ordinary fibromyoma.

The uterine cavity, including the cervical canal, measured four inches in length. The mucosa of the cervical canal presented a number of longitudinal rugæ, and was not thickened; whilst that of the uterine canal was slightly thickened and ragged, but presented no marked polypoid excrescences, hæmorrhagic foci, or other pathological appearances.

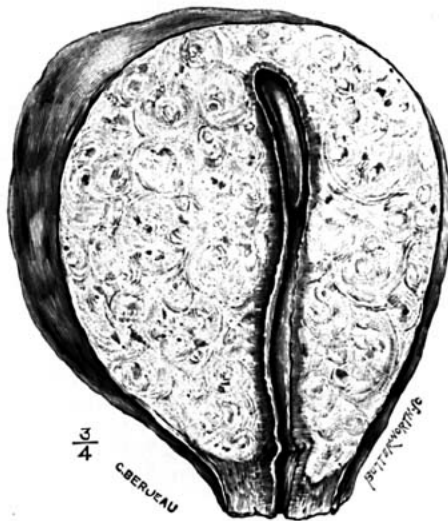
The attached appendage—the right one—showed the tube to be five inches in length, markedly tortuous, distinctly thickened and indurated, especially in its ampullary portion. The abdominal ostium was patent and was surrounded by large spongy fimbriæ. The

ovary measured $1\frac{3}{4}$ in. \times 1in. \times $\frac{3}{4}$ in. It was rough and puckered on its surface, and was white in colour except at one pole where the colour of a hæmorrhagic cyst showed through. On section several small cysts were seen; the largest, the size of a filbert, contained reddish-brown blood clot. The mesosalpinx was thickened so that no trace of the parovarium could be seen. The pedicle formed of broad ligament was also thickened and contained an excess of fibrous tissue and an unduly large number of blood-vessels. The opposite appendage showed very similar changes.

Histological Examination. The endometrium shows no abnormality, being composed of gland tubules embedded in an abundant stroma and covered by a layer of columnar epithelium. The epithelium of the tubules is columnar in shape, and the stroma consists of a richly cytogenic or lymphadenoid connective tissue. The bulk of the tumour is composed of bundles of plain muscular tissue, interspersed amongst which are gland tubules embedded in a mass of richly cytogenic lymphadenoid connective tissue. The gland tubules are composed of a single layer of columnar epithelium lying on a basement membrane which is everywhere intact, and except that some of them are slightly dilated into small cystic spaces and are lined by flattened epithelium, exactly resemble the gland tubules found in the endometrium. The presence of cilia can nowhere be detected on the epithelium of the gland tubules. In some of the cells lining the acini two nuclei apparently occupy a cell, and this resembles closely the condition figured by Mr. Bland-Sutton in his work on tumours when describing the "tubular" variety of cancer occurring in the uterus. The stroma exactly resembles that of the endometrium. It is composed of cells of various shapes, some being spindle cells, others round cells, and others again cells with many branching protoplasmic processes, the latter forming a fine-meshed network in the interspaces of which lie the round and spindle cells. The cell bodies are somewhat indistinct, and take up the stain indifferently, whereas the nuclei are distinct and well stained, stand out well, and lie in close proximity to one another. It is in some places directly continuous with the interglandular stroma of the endometrium, strands of gland-tubules, containing lymphadenoid stroma dipping down from the endometrium amongst and between the muscular bundles of the tumour. In other places the same arrangement exists, though no communication with the endometrium can be observed, the masses or islets of gland-containing lymphadenoid stroma being quite surrounded by muscular tissue. The whole thickness of the tumour



Adenomyoma Uteri Diffusum Benignum.



mass forming the spongy portion of the tumour seen with the naked eye shows this structure, so that gland-tubules are present up to within one-third of an inch from the outer serous covering of the uterus. This external layer of one-third of an inch is composed entirely of plain muscle fibres, and shows no traces of glandular structures.

From a consideration of these appearances we conclude that we have to deal with an adenomyoma of the uterus, the name adenomyoma denoting a new formation composed of glandular and muscular elements. We would distinguish, however, between a new growth due to muscular and adenomatous proliferation, that is to say, an adenomyoma proper, and a myomatous tumour which, in the process of its growth has cut off and included within itself glandular elements, such a tumour being more properly designated a "myoma with glandular inclusions."

In considering the ætiology of this variety of tumour it seems advisable to consider the matter from an embryological standpoint, since von Recklinghausen,* whose exhaustive researches are contained in the classical monograph on the subject, was strongly of opinion that uterine adenomyomata almost exclusively owed their origin to persistence of embryological structures. In his opinion the embryological structures from which these tumours are derived are in nearly every case persistent remnants of the Wolffian ducts, although he allows that in one of his cases the origin of the tumour could not be traced to these structures, but was probably derived from the uterine mucosa. It should be recalled that in early foetal life the Wolffian and Müllerian ducts lie in close proximity to one another from the kidney to the sinus urogenitalis. The Wolffian duct, which persists as Gartner's duct, passes beneath the Fallopian tube in the broad ligament towards the uterus, into the lateral aspect of which it dips opposite the level of the os internum. It then passes through the cervix to the portio vaginalis and thence becomes reflected on to the vaginal fornix, runs along the lateral aspect of the vagina, and finally ends at the free border of the hymen. There is a specially close relationship between the Wolffian and Müllerian ducts at the point where the Wolffian duct passes beneath the Fallopian tube, close to the uterine cornu. It is held by some that the adenomyomata have their origin in the Müllerian ducts, whilst

* Die Adenomyome und Cystadenome der Uterus und Tuben-wandung. Berlin, Hirschwald, 1896.

others, closely following von Recklinghausen, hold that the *fons et origo* are the Wolffian ducts. In support of the latter theory it has been demonstrated by Meyer, Klein and others that "rest cells" derived from the Wolffian ducts occasionally exist in the wall of the uterus underlying the serosa, especially in the neighbourhood of the uterine cornua, and consequently the subperitoneal adenomyomata recognised by von Recklinghausen would seem to have a not unlikely origin from this source. Nevertheless even in this locality adenomatous formations have been found which seem to point to a uterine mucosal origin. The pathological difficulty in connection with the differentiation of tumours from these respective ducts is consequently very great, and it has been suggested that a means of distinguishing them may be found by consideration of the tumour site. That is to say, adenomyomata derived from the Wolffian ducts are subperitoneal and dorsal, whilst those derived from the Müllerian ducts are intramural or submucous and very frequently ventral.

On observing the course of the Wolffian ducts it can be readily understood that adenomyomata may occur in the Fallopian tube, broad ligament, uterus or vagina. The muscular elements in the tumour in such cases would probably be referred to the muscle substance of the uterus, which has been stimulated to increased growth by the proliferation of the Wolffian epithelium, the combined proliferation of muscular and epithelial elements thus giving rise to the formation of an adenomyomatous tumour. A similar stimulation of the musculature of the myometrium may be induced by its invasion by endometrial epithelium in those cases where the tumour is mucosal in origin.

According to the relative proportions of glandular and muscular elements present the consistency of the tumour will vary. Those in which the muscular substance greatly preponderates are hard, those in which there is an excess of glandular structure are soft, while still softer are those of the telangiectatic variety. Occasionally the gland tubules are dilated and contain fluid so that the tumour may attain large dimensions and be quite cystic, forming the variety known as cystadenomata. In accordance with these differences von Recklinghausen classifies the adenomyomata into four varieties:—

1. Hard adenomyomata, where muscle predominates.
2. Soft adenomyomata, where cellular elements are in excess.
3. Softest adenomyomata, or telangiectatic.
4. Cystic adenomyomata, where the gland-tubules are dilated and contain fluid.

The subperitoneal tumours are harder than those which are intra-

mural. The commonest site of all for these adenomyomatous tumours is the tubal angle. They are as a rule intra-mural, but have a strong tendency to become subserous, the serosa being bulged out in flat masses, and in some cases the peritoneum may be perforated.

There is no regular capsule around the tumour margin, and consequently they are of an infiltrating nature, or rather their infiltrating nature allows of no sharp differentiation from surrounding structures. However feasible the Wolffian "rest cell" theory of origin may seem there can be no doubt that many tumours owe their existence to the uterine mucous membrane. As noted above, such tumours are apt to lie centrally and very frequently ventrally, whereas the Wolffian duct adenomyomata have a tendency to be peripheral and dorsal. At the present state of our knowledge there is a certain degree of uncertainty as to the precise mode of origin of some of these adenomyomata. In those tumours arising from the uterine mucosa the mucous membrane can be seen dipping into the muscular substance at various points and penetrating between the muscle bundles. Thus glandular spaces come to lie throughout the tumour surrounded by a rich cellular connective tissue similar in every respect to the tissue found in the normal endometrium. In some recorded cases the presence of cilia has been made out in the cell spaces. The cells are arranged in a single layer, and this arrangement is maintained throughout the tumour mass. The epithelium is of a cylindrical form, but where cystic distension takes place it becomes cuboidal or even flat. In some tumours the cellular investment of the glands is absent at parts, and thus the muscle fibres come into contact with the glands.

The origin from the uterine glands can easily be detected, as at parts these glands are seen to send out offshoots which are accompanied by cellular tissue from the endometrium. These projections at first take the form of a peninsula due to the muscle fibres gradually encroaching on the offshoots, and finally glandular islets exist when the muscle fibres completely separate them from the endometrium. Von Recklinghausen considered this tumour very rare, and of his 33 cases only referred one to this origin. Such has not been the experience of some Continental and American observers. Kossman, Huser, Diesterweg, Lockstädt and Claisse all refer the origin of adenomyomata to the mucous membrane. Another explanation which has received but little attention is that of Eschoff, who considered that the peritoneum covering a myoma might dip into it, and thus be cut off in a manner precisely similar to the mucous membrane canaliculi. This view has been supported

by some of his own observations. Multilocular ovarian cysts may owe their origin to the tubes of Pflüger which are persisting epithelial tubules in the ovarian tissue derived from the germinal epithelium covering the surface of the ovary. Is it not possible that the peritoneum covering the uterus may be modified and having dipped into its substance form adenomyomata by proliferation? In tumours in which there is a tendency towards an increase in number and size of the vessels the telangiectatic variety is formed.

Cullen has described the presence of giant cells, but tubercle bacilli could not be discovered. Some French authorities consider the disease as microbic, and the leucocytosis sometimes found as evidence in support of this theory. The presence of adenomyomata in the broad ligaments has been observed, and they may occur at any part from their free margin to their uterine end. Von Recklinghausen was of the opinion that their occurrence there was purely a secondary matter—the original tumour being formed in the uterine wall and then extruded into the connective tissue of the broad ligament. In a case reported by Pick bilateral and symmetrical tumours existed in the broad ligaments corresponding to the epoöphoron between ovary and tube. Pick, from his own personal observations, differentiates between epoöphoral and paroöphoral adenomyomata—the latter being situated in the mesial portions of the broad ligaments, while the former are in its lateral parts. Many observers (Pick, Cullen, Martin and Cohen) found adenomyomata in the course of the round ligament up to its insertion into the labia majora. Von Recklinghausen frequently described adenomyomata as occurring in conjunction with infantile and malformed uteri. Freund gives the following clinical characters in connection with the adenomyomata:—

1. Patient's age 20—50 years.
2. Generally weak and unhealthy in childhood.
3. Menstruation appears late.
4. They have suffered from anæmia.
5. A constant symptom is dysmenorrhœa with profuse menstruation.
6. The majority are sterile.
7. Pelvi-peritonitic symptoms appear early.
8. Disturbed functions of bladder and rectum.
9. The patient's general health fails.
10. The genitals are infantile in character.

The vagina is short, fornices shallow, uterus anteflexed, cervix small, widening towards fundus.

Moreover these cases are less favourable than ordinary myomata, as castration or ligature of the uterine vessels has no effect. There is also a tendency to carcinomatous degeneration, and, according to some authorities, operation is more difficult.

We give here a table contrasting our case with the above :—

1. Patient's age 49 years.
2. Up till the age of seven years she was considered a delicate child.
3. Periods commenced at the 13th year.
4. Patient was not anæmic before the onset of her illness.
5. At the commencement of menstruation (13th year) and for some years afterwards she suffered much pain at the periods, but of late years she had very little pain. The menstrual periods lasted as a rule for seven days. She did not suffer from excessive loss till three years ago, and the periods were regular. During the last three years she has suffered from severe hæmorrhage at the periods, which were prolonged at times for four or five weeks.
6. She had borne four children, and had two miscarriages (first 16 years ago, fourth 6 years ago).
7. Never suffered from pelvi-peritonitis.
8. No rectal or vesical disturbance.
9. General health had failed gradually since the onset of her illness, till now she is very weak.
10. The genitals are well developed.

The patient since the operation has written to say that she is gaining strength daily, and her colour has much improved.