SACRO-COCCYGEAL TUMOURS.

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I RECENTLY had the good fortune to come across a very typical case of congenital sacral teratoma. I had never seen one previously, and I found its study very interesting. During this Session the Academy has had the opportunity of hearing a paper read by Mr. Pearson on epignathus, and I thought the occasion should not be let pass without bringing before you a short paper on the corresponding tumour which occurs at the anal region. My original intention was to deal only with my own specimen, but on consideration I thought it better to deal with sacro-coccygeal tumours in general. I have found no book which treats the matter fully. Most of the authors dismiss it in a few lines or ignore the subject altogether. The reason for this is probably to be attributed to the rarity of the condition. In searching the *Lancet* I found the last two typical cases were recorded as far back as 1897 and 1895. About this time there was a quarterly journal, entitled "Teratologia," edited by Ballantyne, but it did not run long, owing to insufficient support. While it was in circulation it appears to have stimulated the publication of cases of teratomata, and no doubt it dealt fully with the subject of my paper, but I was unable to secure a copy of the work, even from the publishers. A book of the same nature by St. Hilaire appears to have been in existence in the nineties. This,

also, I failed to obtain. I accordingly had to fall back on rather slender resources for the paper, and this, I trust, will explain many of its shortcomings.

I shall begin by describing the specimen in my possession. The patient from whom the tumour was removed was a boy, aged eleven months, in whom a swelling between the anus and coccyx had been present from birth. This swelling gradually increased in size, but caused no special symptoms except that the fæces were flattened. On examination a semi-fluctuating mass was found, over which the skin was freely movable. The rectum was considerably displaced owing to its presence. Before operation a hypodermic needle was passed into the mass, and 1 c.c. of a slightly opalescent fluid was drawn off. This fluid was alkaline in reaction, gave a marked albumin-reaction, failed to reduce Fehling's solution, and on centrifugalisation showed a little blood. The removal of the tumour was accomplished without difficulty.

I have to express my indebtedness to Sir Joseph Redmond and Mr. A. J. Blayney, under whose care the patient came, for the specimen and for permission to bring forward the case.

Macroscopically the tumour consisted of two distinct portions which were encapsuled. Together they weighed 288 grms. The smaller portion measured 45 × 40 × 10 m.m. It was mainly solid, but some small cysts were visible. The larger part was markedly lobulated and studded with cysts of varying sizes up to 3 c.m. in diameter. It measured 110 × 90 × 70 m.m. The cysts were filled with a mucoid fluid. They were closely packed together. There was no appearance of secondary cyst-formation, and no intra-cystic ingrowths were

observed. A few small systs were seen with their walls coloured dark brown. In places thin plates of bone could be felt.

Microscopically the tumour was seen to be made up of glandular spaces, lined with epithelium, embedded in a fairly well developed connective-tissue stroma. This stroma was made up mainly of fibrous tissue with thin-walled capillaries. In places hæmorrhages into the stroma were seen. Fatty areolar tissue was present in patches. Unstriped muscle was abundant. Pieces of hyaline cartilage were numerous, and plates of bone were also found. Elastic fibres were present. No fully-developed blood-vessels were seen. Nerve tissue resembling cerebral cortex was widespread, and in it were numerous corpora amylacea. Heart-muscle was encountered in two blocks of the tumour. A small pancreas was detected showing ducts and islets of Langerhans. The cystic spaces were lined mostly with columnar epithelium. Much of this epithelium was ciliated, and the association of spaces lined with columnar epithelium groups of mucous glands and islets of hyaline cartilage could only be interpreted as an attempt at the formation of a bronchial tree. No Lieberkuhn's follicles were found, but some of the non-ciliated columnar epithelium had scattered patches of lymphoid cells in its neighbourhood, which was regarded as possibly representing portion of the intestinal tract, though no special arrangement of unstriped muscle was noticed in this situation. Some of the epithelium was flattened or cubical, and presented appearances of a transition into squamous epithelium. It was some time before I got a block showing typical squamous epithelium, and this was in a portion which had to be decalcified owing to the presence of bone. The squamous epithelium lined

cystic spaces into which hairs from hair-follicles projected, and sebaceous and sweat-glands were present in the same region. Here, too, was found a developing tooth, so we really had the anlage of a jaw. Typical squamous epithelium was not found in any other portion of the tumour, and its presence in the section containing bone is interesting in connection with Professor Geddes' recent paper before the Academy on bone formation, in which he maintained that the bone-forming cells were derived from squamous epithelium. The cysts with dark-brown walls showed epithelium studded with amorphous pigment-granules, which gave the micro-chemical reactions for melanin—the cysts, in fact, represented rudimentary eyes.

Such were the appearances presented by this tumour, and they constitute a typical picture—a congenital tumour situated between the rectum and coccyx or sacrum, and showing on section numerous cysts of various sizes and several tissues derived from all three layers of the embryo.

That such a tumour may be found in this situation is, of course, well known, but typical examples of it appear to be met with rarely. Bland Sutton (1) divides tumours occurring in this region into three classes—(1) Tumours of the post-anal gut or mesenteric canal; (2) post-rectal teratomas, which, he says, are very rare, and mostly resemble ovarian dermoids; (3) teratomas of rectum growing from the mucous membrane, in which there may be locks of hair presenting at the rectum. Accepting this classification for the moment it will be seen that the tumour of which I have given details can only fit into the second class. Thus we see that teratomas occurring here are of two types—(*a*) One like the ovarian teratoma with a single large cyst and an insular protuberance containing

many tissues in it; and (*b*) a very much more uncommon form somewhat resembling the cysto-adenoma of the ovary macroscopically.

Perhaps, before discussing these two types of tumours, we might consider the tumours and developmental abnormalities that may occur in the anal region. They are as follows :—

1. A blastoma, such as a lipoma.
2. Conditions such as cystic hygroma.
3. A swelling due to proliferation or distention of a remnant of the neurenteric canal or post-anal gut.
4. A true tumour derived from the embryonic remnants mentioned in the previous heading.
5. Congenital sacral teratomata (post-rectal teratomas of Bland Sutton)—

   (*a*) Rather rare, and like the common ovarian dermoid.

   (*b*) Very rare, and like the extremely unusual, small, many-cysted ovarian teratoma.

6. Cases in which a limb, or fused limbs, project from the anal region.
7. Cases where an attached parasite exists.

The first class may be disposed of briefly—blastomas in this situation are uncommon, but their occurrence has to be considered in making a diagnosis. The second, sixth, and seventh headings do not call for special attention. The third type of case is illustrated by a very typical example recorded by Middeldorpf (2). He found a loop of gut embedded in fat between the rectum and coccyx. The lumen had a fistulous communication with the exterior through which mucous was occasionally passed. At the other end the loop was attached to the wall of the rectum,

but there was no communication between their respective lumina. The wall of this coil of gut was normal, showing Lieberkuhn's follicles, circular and longitudinal muscular coats, and many solitary lymph-follicles, but no serous coat was present. The patient was a girl, aged one year, and the operation was successfully performed by Kraske in 1884. It is hard to say what exactly was Middeldorpf's opinion as to the nature of the condition. He appears to have regarded it as a teratoma, but says he could find no reference to a case in the literature in which gut alone was present. He concludes by saying that it probably arose from the neurenteric canal, and this is the only explanation possible, but in the pathological sense it is not a teratoma—not even a true tumour.

This brings us to the fourth heading, and here we meet with tumours macroscopically resembling those occurring in class 5 (*b*). They consist of glandular spaces lined with columnar epithelium embedded in a connective-tissue stroma. They show nothing else. Such we must regard as adenomas or cysto-adenomas of remnants of the post-anal gut or neurenteric canal. Mr. Seton Pringle (3) read a paper before the Academy a few years ago on a very interesting example of this type of tumour. It occurred in a middle-aged man, and had become malignant, giving rise to metastatic deposits in the inguinal glands. Such tumours are rare, but they are not teratomas. Still the names sacro-coccygeal tumour and congenital sacral tumour, being capable of being applied to both conditions, may give rise to confusion.

We finally reach the fifth heading, that into which the tumour I have shown fits. There are, as already mentioned, two varieties. Skutsch (4) collected details of twenty of these tumours in 1899. With one possible ex-

ception, they corresponded to what we may call the common ovarian dermoid type. Some of them were situated above and some below the levator ani muscle. In one patient two such tumours existed.

So far we have been treading on safe ground, but when we come to explain the origin of these teratomata difficulties occur. Adami (5) regards the more solid type of teratoma as due to redundant growth of the inferior growing-point. Over-activity of the superior growing-point undoubtedly affords a satisfactory explanation of the condition known as epignathus. But in the anal region we do not find an exactly similar condition. In the latter case we have an irregular mass of tissues, not definitely encapsuled, and springing from some bony mass, usually at the top of the pharynx. In the former situation we always find, as far as I am aware, an encapsuled tumour, unless malignant change has supervened. Over-activity of the inferior growing-point does explain cases of reduplication of the external genitals. When the over-activity is symmetrical—that is, when bifurcation occurs—we get such abnormalities as katadidymus tripus. If we accept over-activity of the growing-point as explaining epignathus and congenital sacral teratoma, we are faced with the difficulty of explaining the occurrence of tumours resembling the latter type in such places as the mediastinum, the neck, and the ovary, which are far removed from the growing-points. Probably sacral teratoma (both types) are best explained on the same lines as those found in the ovary, though it must be admitted that these explanations are not quite satisfactory. But granted the explanation of the sacral teratoma, we still do not know why it is at one time a unilocular cystoma and at another time is multi-locular, no more than we know

why a papilloma occurs in one patient, an epithelioma in another. In spite of the great advances made in the pathological anatomy of new growths the most important questions arising out of them remain unsolved.

NOTE.—I have to acknowledge my indebtedness to the Curator of the Library of the College of Physicians for permission to look up some of the literature referred to, and to Professor Bayley Butler, of University College, for the micro-photographs which were shown during the reading of the paper.

REFERENCES.

(1) Bland Sutton. Tumours: Innocent and Malignant. 1911.
(2) Middeldorpf. Virchow's Archiv., Bd., ci., s. 37.
(3) Seton Pringle. Transactions of the Royal Academy of Medicine in Ireland. Vol. XXV. P. 358.
(4) Skutsch. Zeitschrift. für Geb. und Gyn. 1899. xl., s. 353.
(5) Adami. General Pathology. 1910. P. 239.

PROFESSOR O'SULLIVAN suggested that the material described as heart muscle might be voluntary striped muscle. He thought it was the case that in some dermoid cysts of the ovary small solid portions are found which may contain a great variety of different structures.

DR. T. T. O'FARRELL suggested that Dr. O'Kelly should mount some of the sections and present them to the College Museum. He inquired if Dr. O'Kelly had met with parasitic cysts in the region.

DR. BOXWELL was not sure as to the theory about dermoid cysts.

DR. O'KELLY, replying to the remarks, said as to the heart muscle, he did not see anything that he considered distinctive, but he thought the fibres were short. He considered that the ovarian dermoids were really teratomata. He did not know that parasitic cysts were likely to grow in that situation.