A case of post-rectal dermoid, removed by the parasacral route : with remarks on tumours of the sacro-coccygeal region / by Henry Rutherfurd, M.B., F.F.P. & S.G., Assistant Surgeon, Glasgow Royal Infirmary.

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A CASE OF POST-RECTAL DERMOID, REMOVED BY THE PARASACRAL ROUTE: WITH REMARKS ON TUMOURS OF THE SACRO-COCCYGEAL REGION.

BY HENRY RUTHERFURD, M.B., F.F.P. & S.G.

A CASE OF POST-RECTAL DERMOID, REMOVED BY THE PARASACRAL ROUTE: WITH REMARKS ON TUMOURS OF THE SACRO-COCCYGEAL REGION.

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Mrs. B., aged 34, was admitted to Ward 26 of the Glasgow Royal Infirmary on the 27th December, 1899, with what was supposed to be, and what I myself regarded as a fistula in ano of many years' standing. She stated that at the birth of her first and only child, 10 years before, a few days after leaving bed she began to be aware of a hard painful swelling in the right buttock; this was incised by the medical man in attendance and gave escape to a large quantity of dark-coloured material. The opening had continued to discharge almost continuously ever since. Later on, she told us further, that as an infant she had had a swelling in the same situation which also was incised, but which closed after a time.

On examination, there was found between the anus and the right tuber ischii, a warty-looking granulating surface about the size and shape of a haricot bean; from an opening near its middle there escaped on pressure a small quantity of clear glairy fluid. There was some induration about the raw surface, and slight tenderness was noted. A probe passed into the opening travelled upwards and inwards, and met with resistance after about two inches of probe had been swallowed. With a finger in the rectum, the point of the probe could be felt quite outside the mucous membrane of the bowel about three inches from the anal orifice, the impression conveyed

being that some force would be required to bring it into the lumen of the bowel.

On the 8th January, the patient having been put under chloroform, I laid open the sinus in the direction in which the probe travelled, which was backwards and upwards. I then thought I had come to the end of the cavity, one, namely, in the ischio-rectal fossa; and suspecting the nodular mass in the skin to be tubercular, I proceeded to excise it and the walls of the sinus, noting that the probe had not passed within the pelvic fascia. On cutting away the indurated tract behind and above, it was seen that the section was densely fibrous, and from the upper end of the wound there leaked a thin ill-smelling discharge. An incision into the tissue beyond gave escape to this fluid in large quantity, and the opening was found to communicate with a smooth-walled cavity behind and to the right of the rectum, into which the index-finger went its full length between rectum and sacrum. This opening was in the levator ani and the layers of the pelvic fascia; it was enlarged so as to permit of the cavity being wiped out and stuffed.

So far I had no idea of what the nature of the case really was. I was inclined to regard the cavity as due to chronic abscess, possibly connected primarily with the rectum, or possibly due to a pelvic cellulitis starting from the neighbourhood of the cervix uteri. I even thought of a salpingitis. About six weeks later, when the wound had contracted without diminution of the discharge, I re-opened it and scraped the cavity freely. This was followed by as little improvement; and after this I became convinced that I had to do with a definite cyst of some kind, and the idea of a dermoid came to my mind. I had not made any examination of the scrapings, but perhaps the bitter offensive smell had something to do with suggesting the presence of epithelial débris. On 14th March, 1900, with the kind assistance of Mr. H. E. Clark, I operated by Kraske's method, removing the coccyx and right lower angle of the sacrum. After getting through the levator ani, the wall of the cyst was exposed and somewhat tediously dissected out. The dissection was quite delicate on the side next the rectum, and at the last I applied a ligature before cutting it away. The

cyst itself was about the size of a Tangerine orange, and the cavity left, a rather unpromising one to treat. It could not be filled with soft tissues and had to be stuffed, the skin wound being closed only in part.

The patient was kept lying on her face and the utmost care taken to prevent soiling of the dressings, but by the second day there was a rise of temperature and obvious inflammatory symptoms. Frequent dressings had to be resorted to, and after some time, the cavity having become clean, the wound was further closed by suture. Finally, the remaining raw surface was grafted, but it was not till the 19th June that the patient was dismissed.

She has been seen since (April, 1901), with a healed depression to the right of the lower end of the sacrum, but with no material damage to the floor of the pelvis, though on coughing or straining there is a somewhat diffuse bulging of the scar.

Microscopic examination of the cyst wall, which was kindly undertaken by Dr. Workman, Pathologist to the Infirmary, showed a fibrous-tissue structure with striated muscle fibre, no doubt representing portions of the levator ani, lined by a stratified more or less squamous-celled epidermis. In the fibrous tissue were embedded glands of a convoluted tubular structure resembling sweat glands.

Post-rectal dermoids are to be distinguished from rectal dermoids, and both from dermoids of the peritoneal cavity (ovarian for the most part) bursting into the rectum. Secondly, they are to be distinguished from tumours of a more complex structure, peculiar to the same region, the compound cystic sarcomata of Braune (*Die Doppelbildungen und Angeborene Geschwülste der Kreutzbeingegend*, 1861) or thyroid-dermoids of Bland Sutton, whose origin in the post-anal gut or neurenteric canal of the embryo was first propounded by Middeldorpf (Virchow's *Archiv*, Bd. 101, S. 37).

Mr. Bland Sutton refers to three cases of rectal dermoids, and says "a feature of these cases is that the tumours are furnished with long locks of hair, which protrude from the anus and annoy the patient. Like post-rectal dermoids, they sometimes contain teeth." (Port, *Trans. Path. Soc.*, London,

xxi., p. 307; Clutton, *Trans. Path. Soc.*, xxxvii., p. 252; Danzel, *Langenbeck's Archiv*, Bd. xvii., S. 442.) In the last-mentioned case the tumour, which was carefully described, was evidently complex in structure. It was removed from a woman 25 years of age, and is said to have contained brain-substance and bone, besides bearing on its surface hair and a tooth. The patient died three months after the operation, of what is described as a local peritonitis. The operation was done after dilatation of the anus, and was evidently a severe one, contrasting with what is required when such a tumour is pedunculated. The case reminds us that rectal dermoids are not always pedunculated.

"The removal of pedunculated open dermoids of the rectum," says Senn (*Tumours*, 2nd ed., p. 650) "offers no difficulties; on the contrary, the extirpation of perirectal tumours requires often a formidable operation. Usually the difficulties of operative removal are increased by inflammation and suppuration, which render the dissection tedious and difficult." He refers to "a case of post-rectal dermoid which had suppurated and ruptured just below the coccyx. . . . There was found an opening large enough to admit three fingers, lined by skin and leading to a cavity the size of a child's head lined by hairy skin. Decision was against interference . . . the writer could hardly imagine how such a cavity could be made to heal after dissecting out the entire sac." Such a case is narrated in detail by Deahna (*Archiv für Gynaekol.*, Bd. vii., S. 305). The patient was a girl of 18, who suddenly developed a tumour in the lower abdomen with pressure on the rectum and retention of urine. The lower wall of the cyst was felt between the rectum and sacrum, the upper impinged on the abdominal wall between the umbilicus and symphysis. Exploratory puncture per rectum was followed by suppuration and acute distension, resulting in sloughing of the soft parts including vaginal wall and urethra, analogous, as is pointed out by the writer, to what takes place in protracted labour, from pressure of the foetal head. Here also the patient, who was under the care of Czerny, had to be dismissed relieved of the pressure symptoms but with an open cyst cavity.

Trzebitzky (*Wiener Medicin. Wochenschrift*, 1885, p. 393)

reports a case from Mickulicz's clinic (Cracow) where two separate but connected cysts were present—the one presenting externally in the left buttock, of the size of a child's head; the other, the size of an orange, occupying the left half of the pelvis. They contained a thick yellow fluid with epithelial débris and hairs. The upper one was found to have separated the fibres of the levator ani and to be intimately connected with that muscle. The tumours were dissected out and left a cavity in the pelvis more than the size of two fists. In spite of every care and the use of buried sutures the cavity suppurated and healing was slow. Trzebitzky suggests temporary packing of the rectum in order to secure apposition of the anterior wall of such a cavity to the sacrum.

The same writer is inclined to refer such tumours to errors of development, foetal inclusion in the sense of Cohnheim's theory, in connection with the invagination of the surface epithelium which gives rise to the proctodeum, that is to say, the invagination by which the anus is formed.

The theory which invokes aberrant processes in connection with the proctodeum, while plausible in reference to such cases as the one I have described, is restricted in its application, a restriction which is the more marked as we consider the variety in which congenital tumours develop in the region of the sacrum. Errors of the proctodeum may be invoked to explain simple dermoids—tumours lined by stratified squamous epithelium with or without hair or even sweat glands—but can say nothing to the presence of teeth, still less of bone; nor do they in any way explain tumours whose elements conform to the hypoblastic type. Further, it is to be pointed out that the connection of sacral tumours, cystic or otherwise, is in a proportion of cases rather with the spinal cord than with the rectum, and in these the proctodeum theory is still more obviously at fault. In this connection it is worth noting from a clinical point of view that, as Senn points out, "differentiation of sacral dermoids and of spina bifida is often very difficult, and conclusions should be postponed in doubtful cases until an exploratory puncture has demonstrated the character of the contents of the sac."

So Mr. Bland Sutton (*Tumours*, p. 470), "All reported cases of the successful removal of the sac of a spina bifida from

adults should be carefully studied, because in some instances they may have been dermoids." This is the converse of Senn's statement, and I am able to amplify it by the case of a lymph cyst or hygroma removed by me in the Royal Infirmary from the lumbo-sacral region in a boy of ten. The case had been refused operation elsewhere as a spina bifida, but the tumour, which consisted of three principal sacs connected centrally, was quite superficial to the lumbar aponeurosis.

Among the structures leading to the inclusion of surface epidermis and the formation of true dernoids, there falls to be considered the post-anal dimple or foveola retro-analis. For an account of this structure and its relation to the termination of the spinal cord, I would refer to a paper by MM. Tourneux and Hermann in the *Journal de l'Anatomie and de la Physiol.*, vol. xxiii., "Sur la persistance de vestiges médullaires coccygiens pendant toute la période foetal chez l'homme, et sur le rôle de ces vestiges sur la production des tumeurs sacro-coccygiennes congénitales." Their researches have led these writers to the conclusion that the formation of the post-anal dimple is due to the comparative ascent of the lower termination of the spinal cord caused by the disproportionate growth in length of the spinal cord and the spinal column, and consequent dragging in of the soft tissues with which in its early embryonic stages the cord is in immediate relation. More than one such puckering of the surface may be present, and there may result merely funnel-shaped depressions, or cysts more or less completely shut off from the surface.

Professor Cleland (*Journal of Anat. and Phys.*, Vol. xvii., p. 290) calls attention to the co-existence in cases of well marked dimple of a straight and unduly short sacrum, and is of opinion that both the dimple and the deformity of the sacrum are to be related to an arrested development of the notochord.

The origin of sacral tumours is further complicated by the cases in which we have to do with an unmistakable adherent foetus, and which shade off into those where we seem to recognise more or less well-defined parts, organs or tissues, belonging to a second foetus; and this complexity is increased

by the occasional occurrence of malignancy in some of these tumours, taking the form of sarcomatous or carcinomatous growth with metastases.

Hennig, in Ziegler's *Beiträge*, Bd. xxviii., Hft. 3, gives a valuable review of the theories which have been propounded as to the origin of congenital tumours of this region. Middeldorpf, Ritschl, and Nasse he cites as the exponents of the post-anal gut theory, to whom we must add in this country Mr. Bland Sutton (*Tumours* and *Dermoids*). Hennig himself is of the opinion set forth by Förster, Virchow, and Ahlfeldt, which ascribes to a foetal implantation almost all tumours of the sacro-coccygeal region, which view has also been propounded by Braune (*Die Doppelbildungen und Angeborene Geschwülste der Kreutzbeingegend*, 1861), and in a Paris thesis by Calbet (1893). The summing up of the last-mentioned author is, that the majority of congenital tumours of the sacro-coccygeal region, which are known in literature under the most varied names, belong to one and the same group, and are to be regarded as dwindled (*verkümmerte*) parasitic embryonic deposits (*Anlagen*).

These tumours, which have all a more or less uniform character, possess mostly a very complicated structure. They contain the most various tissues, and may present complete organs or fragments of organs which take their origin in a new foetus to a certain extent grafted on the first, or, as it may be otherwise put, incompletely budded from the first.

The other group of congenital neoplasms of the sacro-coccygeal region, according to Calbet and Hennig, comprises those formations which are to be referred to a disturbance of development, development of a particular structure being in the direction of (1) excess or (2) defect (restriction). To the former class belong the cases of tails, with actual bony vertebrae or of mere tail-like structures, such as have more often been described; to the latter (those due to restricted development) belong all cases of spina bifida, different kinds of dermoid cysts due to the driving-in and cutting-off of processes of the external skin, fistulae, etc.

Reverting once more to the subject of the post-anal gut and its relation to this group of thyroid-dermoids or compound

cystic sarcomata, I desire, while recognising the importance of the observations which concern that structure, to dissent from the suggestion put forward by Mr. Bland Sutton, as to its being represented in the adult by the coccygeal body, or Luschka's gland, and the consequent inclusion of Luschka's gland as an originator of this group of sacral tumours. Hennig (*l.c.*) quotes Virchow as having, as far back as 1861, recognised that Luschka's gland had been found co-existing independently in the presence of the tumours in question. Further, while disclaiming any original anatomical observations on the subject, I think it is open to doubt whether what is known of Luschka's gland (cf. the account of it given in Quain's *Anatomy*) entitles it to be regarded as a structure of epithelial character—that is, of epiblastic or hypoblastic origin.