

**COCCYGEAL SINUS.**

BY R. L. NEWELL, MANCHESTER.

I HAD some difficulty in choosing a title for this paper. Although coccygeal sinus or fistula is the name by which the condition is known in English literature, it is an inaccurate description because the only connection the sinus has with the coccyx is solely one of position. 'Congenital sinuses in the sacrococcygeal region' is perhaps a better title. In America Hodges<sup>31</sup> as long ago as 1880 manufactured the name of 'pilonidal sinus', or nest of hairs.

The condition has received very little attention by British surgeons. Goodsall and Miles<sup>1</sup> in their book on *Diseases of the Anus and Rectum* devote a whole chapter to a condition which they call 'sinus over the sacrum or coccyx', but they fail to recognize its true nature, and attribute its cause to injury. They recognize, however, that it has no connection with the coccyx or sacrum. Even as late as 1932 W. E. Miles<sup>21</sup> still regards it as due to a hæmatoma resulting from a fall, and advances the theory that right-handed people fall more heavily on the left side, accounting for the fact that these sinuses appear to originate on the left side of the median raphé.

Lockhart Mummery<sup>2</sup> drew attention to the congenital origin of the condition in 1921 and demonstrated that the sinuses were lined by epithelium, and later in 1929 published an excellent microphotograph in the *Proceedings of the Royal Society of Medicine*.<sup>3</sup> Towards the end of 1932 W. B. Gabriel<sup>24</sup> published his book *The Principles and Practice of Rectal Surgery*; in it he described the results of his treatment in 9 cases.

It is a condition, however, which has received considerably more attention in America. Harvey B. Stone<sup>4</sup> reports 61 collected cases. P. B. Cattell and L. W. Stoller<sup>6</sup> report 59 cases from the Lahey Clinic, and Frank Glenn<sup>23</sup> reports 120 cases. These figures give the impression that its occurrence is far commoner in America than it is in this country. Personally I am of the opinion that this is not the case, and that the lack of more frequent reference to it in our surgical literature is due to the failure on the part of the practitioner or surgeon to appreciate the pathological basis of the condition. Failure to understand the true nature of the sinus may lead to the mistaken diagnosis of fistula in ano, tuberculous sinus, or even a simple abscess or boil.

I do not doubt that there is hardly a surgeon in this country who has not seen or operated upon several of these cases, and possibly been disappointed with the result of his surgical intervention. P. B. Cattell and L. W. Stoller<sup>6</sup> have followed up 40 cases, and in 9 have found recurrences: 20 out of 50 of their cases had had previous operations.

## SYMPTOMS.

The lesion seldom presents itself to the surgeon until the complication of septic infection has supervened. The following is a typical history.

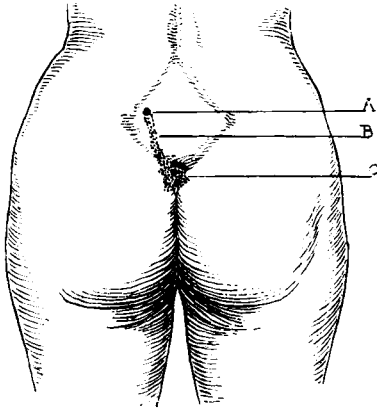


FIG. 137.—Case 1. A diagrammatic representation.

temporarily healed over or showing a small opening with heaped up granulation tissue (*Fig. 137, A*). On palpation a firm hard core is felt leading downwards and towards the middle line (*Fig. 137 B*). In this position can be seen one or several tiny orifices situated accurately in the middle line (*Fig. 137, C*). These orifices may be situated quite close together or may be separated by a distance of as much as an inch. On pressure over the sinus at A and B (*Fig. 137*) pus may sometimes be seen to exude from the orifices in the median raphe.

If the sinus is injected at A (*Fig. 137*) with lipiodol, the solution will, after having filled up the whole of the track, be seen to exude from the orifices in the median raphe (*Fig. 137, C*). *Fig. 138* shows an X-ray photograph after a lipiodol injection.

At an age of from 20 to 24 years a small abscess appears to one side of the middle line (*Fig. 137, A*). This is either incised or discharges spontaneously. After a few days the discharge ceases, but a small thickened nodule remains. The amount of pus is usually very small, and only rarely does the sinus discharge profusely. After an interval of a few days or weeks, this nodule again becomes painful, swells, and discharges pus. Sometimes the condition may remain quiescent for many months. This state of affairs goes on for an indefinite time until the condition is correctly diagnosed and the underlying cause removed.

On examination we find a small hard nodule to one side of the middle line, either temporarily healed over or showing a small opening with heaped up granulation tissue (*Fig. 137, A*). On palpation a firm hard core is felt leading

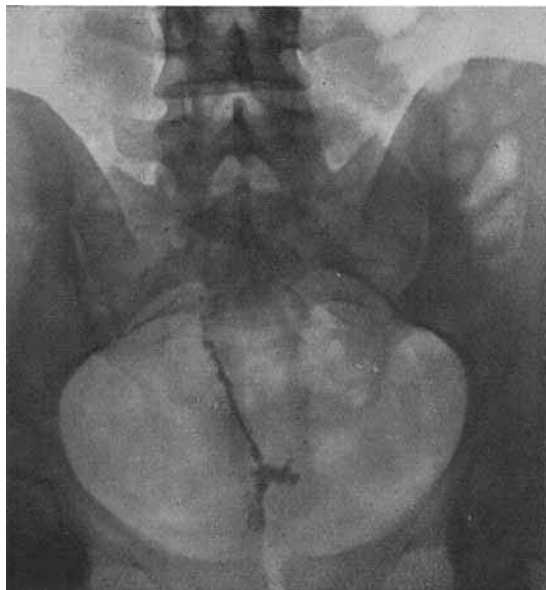


FIG. 138.—Case 1. X-ray photograph showing the sinus after a lipiodol injection. The upper narrow portion is the secondary sinus, the lower expanded portion the primary sinus in the median raphe.

The whole of the track is well outlined from A to the expanded cavity at C (*Fig. 137*). The narrow projection downwards below the expanded cavity is due to some of the lipiodol having escaped on to the surface of the skin.

**HISTOLOGY.**

A microscopic examination of the whole sinus track reveals the following appearance. The expanded cavity C (*Fig. 137*), which is the origin of the condition, is a cavity partially lined by true skin and may contain quite

FIG. 139.—Case 1. Wax cast of the whole of the sinus. The thin portion to the left represents the secondary sinus. The expanded portion to the right, to which hairs can be seen adhering, represents the primary sinus.

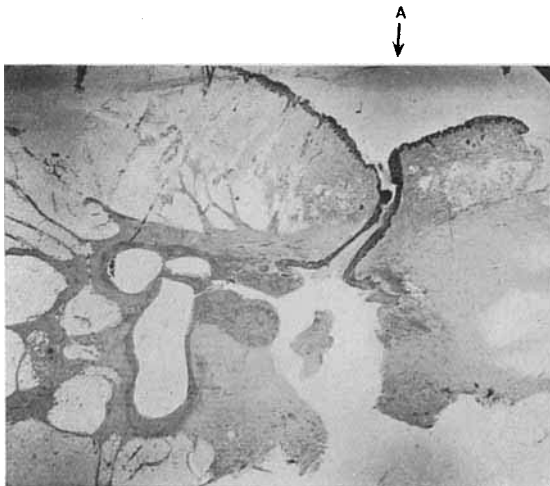
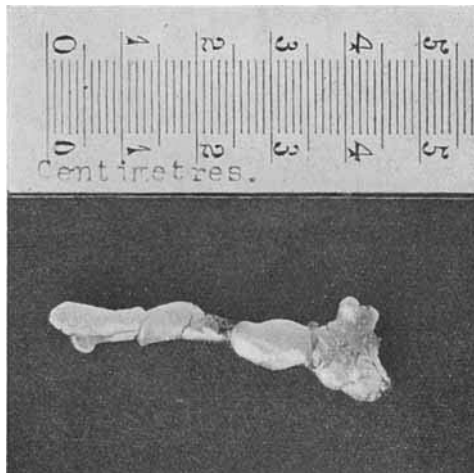


FIG. 140.—A horizontal section taken to show the connection of the primary sinus with the skin and deep tissue. A, Tiny orifice situated in the median raphe; B, Skin surface; C, Primary sinus lined by squamous epithelium; D, Epithelium expands into a cavity; E, Secondary sinuses lined by granulation tissue.

long hairs. Some hairs can still be seen adhering to the bottom of the wax cast seen in *Fig. 139*. This cavity communicates with the surface on the median raphe by orifices lined by true skin. These facts are well illustrated

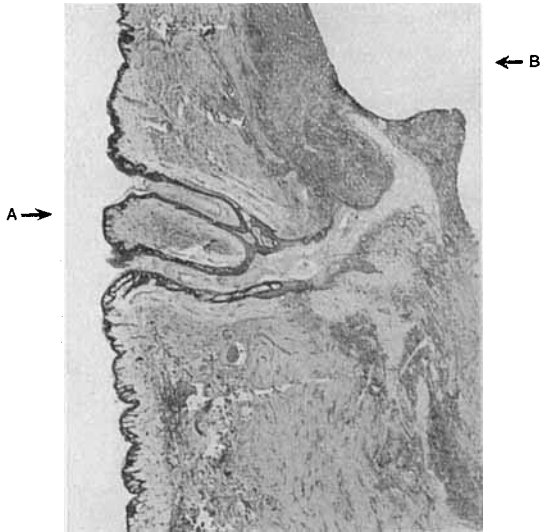


FIG. 141.—A section of specimen shown in *Fig. 147* taken in the mesial plane. It shows two primary sinuses in the median raphe, one above the other. These two sinuses join and extend backwards towards the coccyx. The epithelium appears to be intact. The secondary sinus was caused by one of the other two primary sinuses present in this case. A, Two small primary sinuses in the median raphe; B, Secondary sinus.

FIG. 142.—A vertical section in the coronal plane at right angles to *Fig. 140*.

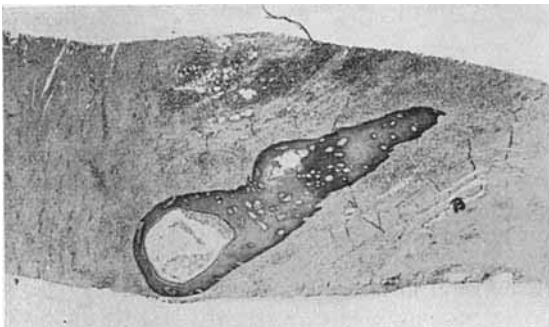
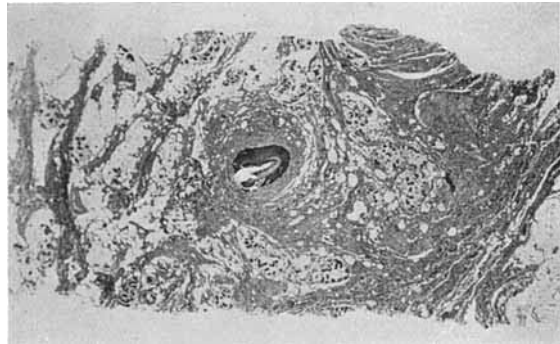


FIG. 143.—A vertical section nearer the coccyx than *Fig. 142*. The sinus is now expanding into a small cavity.

by the microphotographs (*Figs. 140-144*). The sinus B (*Fig. 137*) is purely a secondary sinus and is lined only by granulation tissue. This accounts for the fact that the pathologist, on examining the tissue removed, will often send back a report that it is septic granulation tissue.

### ETIOLOGY.

The exact etiology of the condition has not yet been proved conclusively, but it probably results from change in embryonic development. Harvey B. Stone<sup>4</sup> gives to J. M. Warren<sup>7</sup> the credit of first describing the condition in 1867. Warren regarded a reversed hair follicle as being the cause of the trouble, and as the hair continues to grow "it pulls its hole in after it."

Wendelstadt<sup>8</sup> advances the following theory. The inferior end of the spinal cord is the last portion to lose its connection with the skin. As the sacrum closes together it pinches off this connection, and the skin becomes attached by fibrous tissue at this point to the posterior surface of the sacrum. As adult life approaches, fat and soft parts grow and lift the skin farther away from the bone, and the attached spot is drawn down into a dimple or sinus. P. B. Cattell and L. W. Stoller<sup>6</sup> give Oehlecker's<sup>9</sup> views without comment. He believes that these sinuses are caused by the traction of the caudal ligament on the median raphé. Hermann and Tourneaux<sup>22</sup> regarded the condition as due to a vestigial remnant of the spinal cord. They state that after the separation of the spinal cord from the superficial tissues a process of epithelial tubules persists for a time at the point of previous connection of the neural axis with the skin, forming a vestigial remnant of that connection.

Harvey B. Stone,<sup>5</sup> in a more recent paper, suggests as a possible cause that the sinuses are analogous to the preen glands found in a great many species of birds. The preen gland lies embedded in fat over the last caudal vertebra. It consists of numerous straight tubules lined by polyhedral cells. The tubules converge upon a collecting chamber or cavity, which in turn empties into an epithelial-lined duct opening on to the skin of the back. Their function is either that of oiling feathers or it is sexual.

Ripley<sup>10</sup> and Moise<sup>11</sup> both regard the condition as analogous to spina bifida, and have recorded two cases of staphylococcic meningitis resulting from a congenital sacral sinus. In both their cases there was, however, a direct communication with the spinal meninges and a maldevelopment of the vertebral bodies. Bland-Sutton<sup>12</sup> considers that the condition is due to faulty coalescence of the cutaneous covering of the back, and mentions as analogous the interdigital pouch of the sheep, which often gives trouble if



FIG. 144. — High-power view of *Fig. 143*, showing a cavity containing hairs and epithelial débris and lined by epithelium.

the orifice becomes occluded. He says that they are sinuses frequently associated with lumbo-sacral spina bifida. This statement, however, is not corroborated by an analysis of the published cases.

It would appear, then, that we have to consider two main theories:—

1. That the condition is a true sequestration dermoid, due to faulty coalescence of the middle line.

2. That the condition arises from that remnant of the spinal canal known as the coccygeal vestige.

I do not think the condition is entirely analogous to a true sequestration dermoid. Coccygeal fistulæ are comparatively frequent, and are in a constant position, whereas dermoid fistulæ arising in the middle line are very rare in other parts of the body. I therefore consider that the area in which they are found has a definite influence on their etiology.

As regards the theory that they arise from the coccygeal vestige, we have the work of Hermann and Tourneaux<sup>22</sup> to support this hypothesis. They showed that in the fœtus at a definite stage there is a connection of the coccygeal vestige with the skin. On the other hand, recent work by Kunitomo,<sup>28</sup> who had available for his research the unrivalled collection of embryos at the Carnegie Institute, has failed to confirm this statement. Kunitomo states, "The caudal end of the coccygeal medullary vestige appears to adhere to the epidermis, but in reality it does not."

What, however, is very significant in his work is that he found the various stages in the reduction of the tail bud, as shown on the skin, do not present the same appearance in every embryo, but, on section, evidence of its reduction and disappearance are invariably found dorsal to the caudal end of the vertebral column and in the median line of the embryo.

A coccygeal fistula is lined by tissue indistinguishable from true skin. If it arose from the coccygeal vestige, the cells lining it would have become so differentiated as to be quite unlike true skin. In microscopic sections of the condition there is absolutely no sign of any other tissue than that normally contained in the skin.

I venture to suggest that the following is the true origin of the condition. The sinus is a dermoid fistula, but differing from a true sequestration dermoid in that it is caused by traction of the underlying tissue upon the median raphé. This traction is probably caused by the retrogression of the tail bud. It therefore could be described as a traction 'dermoid'. It may be suggested that the following is the probable sequence of events. The epithelial-lined fistula is present at birth. It would appear to remain a considerable time without giving rise to symptoms. However, owing to the growth of the body and consequent enlargement of the buttocks, the secretion of this epithelium is unable to escape by the orifices in the middle line. These orifices are in a position which is extremely liable to sepsis, being situated at the bottom of a fairly deep cleft. The epithelial fistula becomes infected, and the formation of an abscess follows. This abscess disintegrates a portion of the epithelial lining and a sinus track is formed which tends to extend along the line of least resistance, which is upwards and outwards. This secondary sinus eventually opens on to the surface to one side of the middle line.

## CASE REPORTS.

The following cases have been operated upon by me by the method I am about to describe. In all cases the condition appears to have been completely cured.

*Case 1*—M. C., female, aged 26. The sinus had been discharging for twelve months. There were three primary sinuses in the middle line and one secondary sinus opening on the left of the middle line. Operation on Feb. 5, 1932. The wound became septic and healed by granulation tissue.

*Case 2*—C. J., female, aged 30. The sinus had been discharging for two years. She gave a history of one previous operation. There were two primary sinuses in the middle line and one secondary sinus opening on the left of the middle line. Operation on Feb. 10, 1931. Wound healed by first intention.

*Case 3*—G. A., female, aged 37. The sinus had been discharging for two years. She gave a history of one previous operation. There were three primary sinuses in the middle line and one secondary sinus opening on the right of the middle line. Operation on July 26, 1932. The wound became septic and healed by granulation tissue.

*Case 4*—M. P., female, aged 20. The sinus had been discharging for two and a half years. She gave a history of one previous operation. There was one primary sinus only in the middle line. Operation on Sept. 29, 1932. The wound healed by first intention.

*Case 5*—E. H., female, aged 25. The sinus had been discharging for ten years. She gave a history of three previous operations. There were two primary sinuses in the middle line and one secondary sinus opening on the left of the middle line. Operation on May 31, 1932. The wound became septic and healed by granulation tissue.

*Case 6*—H. P., female, aged 25. The sinus had been discharging for four years. She gave a history of one previous operation. There were two primary sinuses in the middle line and one secondary sinus on the left of the middle line. Operation on Nov. 4, 1931. The wound became septic and healed by granulation tissue.

*Case 7*—F. H., female, aged 30. The sinus had been discharging for six months. There was one primary sinus in the middle line and one secondary sinus to the left of the middle line. Operation on Feb. 8, 1933. The wound became septic and healed by granulation tissue.

*Case 8*—E. W., female, aged 32. The sinus had been discharging for twelve years. She gave a history of two previous operations. There were four primary sinuses in the middle line and one secondary sinus on the left of the middle line. Operation on Feb. 10, 1933. The wound became septic and healed by granulation tissue.

*Case 9*—K. B., female, aged 23. The sinus had been discharging for two and a half years. She gave a history of several incisions. There were four primary sinuses in the middle line and one secondary sinus on the left of the middle line. Operation on Feb. 14, 1933. The wound became septic and healed by granulation tissue.

*Case 10*—M. B., female, aged 23. The sinus had been discharging for one year. She gave a history of several incisions. There were two primary sinuses in the middle line and one secondary sinus on the left of the middle line. Operation on Jan. 31, 1928. The wound became septic and healed by granulation tissue.

*Case 11*—A. S., female, aged 20. The sinus had been discharging for two years. She gave a history of two previous incisions. There was one primary sinus in the middle line and evidence of a healed secondary sinus to the left of the middle line. Operation on March 17, 1933. The wound became septic, and is healing by granulation tissue at the time of writing.

All these cases occurred in women. Frank Glenn's figures show, however, that it occurs more frequently in men. There were 23 females and 97 males in his 120 collected cases. The earliest onset of symptoms was at the age of 15 years, and the latest onset at the age of 35 years.

All these cases were septic at the time of operation, and primary healing was obtained in only two of the series. The remainder healed by granulation tissue. The time taken for complete healing varied from three to ten weeks. In no case have the symptoms recurred.

### TREATMENT.

Although successful treatment has been reported with non-surgical methods by Maillard<sup>13</sup> and Crookall,<sup>14</sup> it would appear that the only satisfactory way of curing the condition permanently is by surgical excision. A complete radical excision which removes the whole of the area involved is absolutely necessary, otherwise recurrence is inevitable. Failure to undertake a preliminary investigation of these cases with a view to obtaining a true diagnosis and an accurate delimitation of the sinus results in many of the patients having to undergo several operations before obtaining a cure. The following method has been found to give very satisfactory results:—

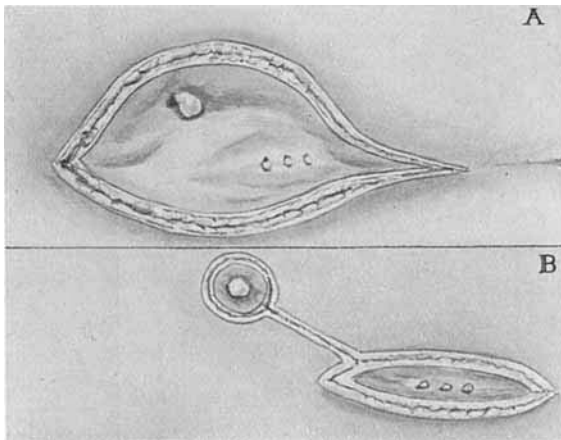


FIG. 145.—A, The incision used when the secondary sinus opening is near the openings of the primary sinuses; B, The incision used when the secondary sinus opening is situated some distance away from the openings of the primary sinuses.

Immediately before the operation, when the patient is under the anaesthetic, the opening of the secondary sinus (*Fig. 137, A*) is injected with melted paraffin wax. Coloured wax can be employed, but there is no advantage in this, as the wax is only used to make the whole of the sinus palpable and to serve as an indication if the sinus is cut across during the operation. A circular incision is now made around the orifice of the secondary sinus, and a straight incision directly over the underlying secondary sinus is made downwards towards the middle line (*Fig. 145B*). That portion of the median

The secondary sinus opening (*Fig. 137, A*) is injected with lipiodol. A blunt needle of fairly wide bore is used so as to fit tightly in the opening. It will be noticed on injecting the fluid that the lipiodol will begin exuding from the tiny orifices in the middle line (*Fig. 137, C*). Excess of lipiodol must be carefully wiped away from the cleft between the buttocks. An X-ray photograph is now taken without disturbing the patient. This photograph gives a fairly accurate idea of the extent of the condition (*see Fig. 138*).



raphé which is affected is now excised by an elliptical incision. If the secondary sinus opening lies near the middle line, then the incision must include both primary and secondary openings (*Fig. 145A*). The incision in *Fig. 145B* will, however, facilitate the closure of the wound. The skin-flaps are dissected well back, taking care to keep fairly superficial (*Fig. 146*). The whole of the sinus track is now removed (*Fig. 147*), beginning at the upper portion. There is no need to make a wide excision of the track from A to B (*Fig. 137*), as the track in this situation is lined only by granulation tissue and has no epithelium lining its walls. The condition, however, is different when the middle line is approached. Here we are dealing with the primary sinus, which has an epithelial lining, and it is advisable to excise not only the sinus but also the skin of the median raphé

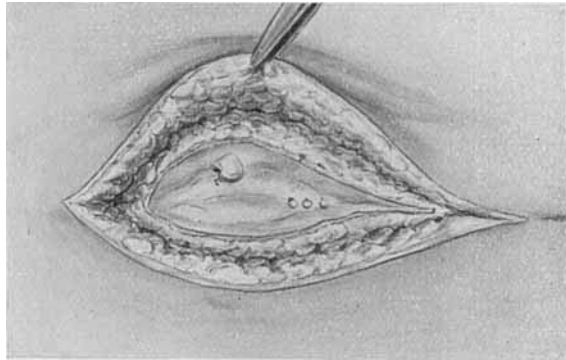


FIG. 146.—The skin edge is undermined.

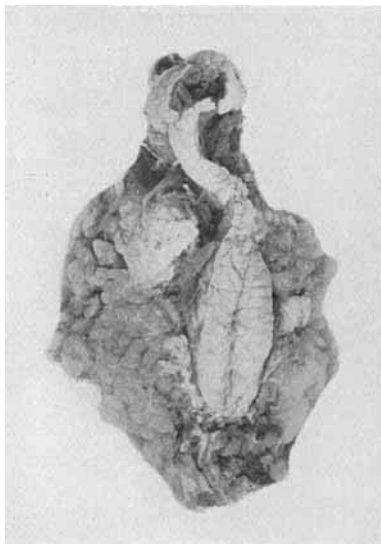


FIG. 147.—*Case 8*. Photograph of the sinus removed. The lower portion of skin shows four small openings in the median raphé. The narrow portion of skin passing upwards and to the left is overlying the secondary sinus. The opening of the secondary sinus can be seen at the top of the specimen.

and a generous portion of tissue around the sinus. Otherwise it is possible that any pits or depressions of the epithelium lining may be cut across and left behind.

This dissection will, of course, leave a fairly large cavity extending forwards to the coccyx, and an attempt must be made to obliterate this cavity as far as possible by undermining the fat and approximating it with catgut sutures. The skin is then sutured without drainage. An application of new skin helps to prevent contamination of the wound. If the wound becomes septic, as is not improbable, then the sutures must be removed and the wound allowed to heal by granulation tissue. Irrigation with a eusol solution should be performed twice daily, and later light packing with an allantoin solution will be found very efficacious in stimulating granulation tissue formation. Every effort should be made to obtain primary healing by swabbing the cavity with B.I.P.P., by undermining the fat, and by obliteration of the cavity with as little tension as possible by means

of catgut sutures through the fat. If primary healing is not obtained, it may be as long as two months before healing by granulation tissue takes place.

### CONCLUSIONS.

1. A coccygeal sinus must be regarded as due to a defect in embryonic development. It probably is the result of traction on the skin caused by retrogression of the tail bud.

2. The treatment consists in the removal of the whole of the sinus, together with that portion of the median raphé which contains the origin of the condition.

3. The extent of the sinus may be difficult to recognize without a lipiodol injection, followed by an X-ray examination.

4. The dissection is rendered easier by an injection of paraffin wax immediately prior to operation.

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