THE ORIGIN OF SACROCOCCYGEAL PILONIDAL SINUSES BASED ON AN ANALYSIS OF FOUR HUNDRED SIXTY-THREE CASES*

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Pilonidal sinuses, with associated cysts or abscesses, have been recognized for over 100 years; however, the recent war served to make physicians more aware of the prevalence and the importance of this condition. The articles written during the war years (1942–48) nearly equalled in number all that had appeared previously. Most of these papers were concerned with treatment and presented conflicting opinions as to the proper surgical procedure. This, however, has not been the only area of confusion. There has been no unanimity of opinion as to origin and pathogenesis. Most writers have agreed that these lesions arise upon a congenital basis but have disagreed upon the pathogenesis. Recent reports of similar sinuses in other regions have revived interest in earlier suggestions of an acquired rather than a congenital origin for sacrococcygeal pilonidal sinuses.

In 1946 and 1948 Patey and Scarff^{1,2} described a lesion on a barber's hand with histopathologic findings identical with those of a pilonidal sinus. They concluded that pilonidal sinuses could not be explained fully on the usually accepted developmental basis but considered them to be acquired infective and foreign body granulomatous reactions to buried hair. In 1947, King³ also suggested that pilonidal sinuses and cysts were results of infection and foreign body granulomatous reaction to hairs which were introduced into the sinuses from without. The example recorded by Ewing⁴ in the same year was less convincing to that author. Recently, Downing⁵ has described another example of pilonidal sinus of the hand in a barber who had expressed hair shafts repeatedly from a lesion of the right second interdigital space, an area in which hair follicles do not occur normally. Apparently, interdigital pilonidal sinus may be considered an occupational disease of barbers.

Examination of such reports of pilonidal lesions in other than the usual area led me to reconsider the problem of the pathogenesis of this condition. There was a significant implication that the essential basis was acquired rather than developmental. Accordingly, a comprehensive review of the literature as to pathogenesis was undertaken and

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pertinent information was extracted from an unselected series of 463 cases on which microscopic sections were available.

HISTORICAL REVIEW WITH REFERENCE TO ORIGIN AND PATHOGENESIS

Anderson,⁶ in 1847, apparently was the first to describe the condition now known as pilonidal sinus, in a report on *Hair Extracted from an Ulcer*. In 1854, Warren⁷ gave a good description of such lesions and reported 2 cases. Later,⁸ adding another case, he suggested a theory as to origin and a method of treatment. He believed that the condition began in a single follicle from a hair which became inverted and continued to grow, forming a tangled mass. Constant pressure and moisture of the area "softened both the newly formed hair and epidermis cells surrounding the mouth of the follicle."

Hodges⁹ was one of the first to consider a congenital basis in the pathogenesis of pilonidal sinuses. He believed three factors to be essential to their development: a congenital coccygeal dimple, abundant development of hair in the area, and poor local hygiene.

Lannelongue ¹⁰ demonstrated that the skin over the coccyx is tightly adherent in the embryo, and that, as the surrounding mesoblastic structures increase in thickness, the natural result would be for the skin to be pulled in at this point, forming a dimple. Any exaggeration of this dimple would cause the formation of a sinus. Then, if the epithelium over the external opening proliferated, it would occlude the mouth, forming a cyst. Since epidermal epithelium lined the cyst, normal skin appendages could be found in the cyst.

Tourneux and Herrmann¹¹ (1887) believed that the embryonic neural canal remains attached to the skin. As the fetus develops, the spine grows more rapidly than the surrounding tissues, and, as a result, the end pulls on the skin, forming a U-shaped epithelial cord around the tip of the coccyx. Normally, this portion of the cord atrophies and disappears after the fifth fetal month; however, occasionally it remains. Since it is composed of stratified squamous epithelium, it may develop a cyst or sinus containing the accessory dermal structures such as hairs and sebaceous and sweat glands.

A few years earlier, Fére¹² had stated, without elaboration, that pilonidal sinuses were slight defects in the coalescence of the superficial portions of the medullary folds in the sacrococcygeal region. This theory, which explained pilonidal sinuses on the basis of incomplete fusion of the lateral halves of the dorsum of the fetus, gained numerous adherents, including Sutton.¹³

These basic hypotheses have been supported and somewhat modified by subsequent workers. Mallory¹⁴ examined the caudal end of seven fetuses, 3 to 6 months old, and concluded that obliteration of the medullary canal takes place first and most completely at the level of the lower end of the sacrum and extends from this point in both directions. He thought that pilonidal sinuses probably originated through incomplete obliteration of a previously existing canal; and, since these lesions extend upward and posteriorly, the medullary canal seemed the most likely place of origin.

Bookman¹⁵ concluded that pilonidal sinuses were due to displacement of "dermal or dermoid" cells in the embryo during the process of fusion in the midline, with these cells assuming an abnormal location under the skin. Collection of epidermal products, as hair and sebaceous material, results in the formation of a cyst.

Stone 16,17 stated that pilonidal sinuses must be regarded as special downgrowths of epithelium originating from the skin. He suggested that they may be a phylogenetic representation of the preen gland which is found in some species of birds. After detailed study of the human embryo, Fox18 concluded that pilonidal sinuses are derivatives of cutaneous ectoderm which has persisted after the period of normal ectodermal invagination has been completed. He stated that an analogy can be drawn, because of this mode of origin, between pilonidal sinuses and a special "scent" gland in the sacrococcygeal region of birds and amniotes. To him this suggested that they might represent a vestigial skin appendage developing at puberty, thus explaining the age incidence of the lesion. Somewhat along a similar vein were the conclusions of Kallet.19 He insisted that the concept that in the sacrococcygeal region there are present at birth embryonic remnants of a vestigial secondary sex gland which is activated at adolescence by the pituitary body better explains the clinical picture than the previously mentioned points of view.

Of more recent writers, Gage²⁰ was the chief proponent of the "neurogenic" theory, *i.e.*, that pilonidal sinuses are due to persistence of the neurenteric or neural canal. According to this view, normally, in the caudal end of the embryo, the portion of the neural canal that is formed by the neural folds and lies between the skin and the coccygeal vertebrae is obliterated by cohesion of its walls. Failure to do so forms a cavity which may be connected to the skin by one or more sinuses. The lining of the cavity retains its capacity to produce primitive skin appendages and a rudimentary type of nervous tissue, if a connection with the spinal canal remains. He believed that the sacral dimple is the result of an anterior pull on the overlying skin by the caudal ligament as the coccyx grows downward and curves anteriorly. This latter idea on the origin of the sacral dimple was first described

by Oehlecker²¹ when he called attention to the relative absence of hair in the area, naming it the "sacral bald spot."

Most subsequent workers are of the opinion that pilonidal sinuses are congenital in origin, and are proponents of either the ectodermal invagination theory or the neurogenic theory.

It was not until 1880 that the name "pilonidal" (pilus: a hair + nidus: a nest) was given to the lesion by Hodges. Since then a variety of names have been applied. Some of the more common names are: sacral, coccygeal or sacrococcygeal infundibulum; dermoid, and dermoid fistula, sinus, or cyst; posterior umbilicus; post-anal dermoid; congenital dermal sinus, and sacrococcygeal ectodermal sinus. However, pilonidal sinus is now the term most generally used.

STRUCTURAL FEATURES OF SIGNIFICANCE IN PATHOGENESIS

The sacrococcygeal pilonidal sinus consists of an acute or chronic inflammation of a localized area. A small opening is seen usually in the midline about 3.5 to 5 cm. posterior to the anal orifice, between, and usually concealed by, the buttocks. This opening, from which hairs may protrude, leads into an epithelium-lined sinus which is directed upward. There may be a bulbous ending to this single tract; or there may be a number of sacculations communicating with each other by way of the main tract or lateral tracts; or some of these may communicate with the skin through other orifices. These sacculations vary in size and are usually filled with vascular pyogenic granulation tissue.

By 1946, sixteen lesions resembling pilonidal sinuses had been reported which had dural connections in direct continuity with the filum terminale.²² Two of these were associated with spina bifida occulta,^{23,24} and two were associated with meningitis.^{25,26} These cases are believed to be examples of congenital anomalies and not of true pilonidal sinuses. If Gage²⁰ correctly recognized glial tissue beneath the epithelial lining, his case, also, must have been a congenital anomaly.

The presence of cutaneous appendages, as hair follicles, sebaceous glands and sweat glands (described first by Gussenbauer²⁷ in 1893, and by Crone²⁸ in 1917), as part of the epithelial lining has been asserted by various writers including Stone^{16,17} and Rogers and Hall.²⁹ Others have not been able to confirm these findings.^{1,3,21,30} They have found areas of acute or chronic inflammation with numerous foreign body giant cells, some hairs without hair follicles, small islands of squamous cells either free or attached to the lining of the sinus, and squamous epithelium at the entrance of the sinuses. Oehlecker²¹ made careful microscopic studies (including serial sections) but found no epithelial structures. It is difficult to explain the conflict of opinions on the pres-

ence of hair follicles and other accessory skin structures. This is a matter of objective observation and should have only one answer. Does it mean that those who claimed the presence of such structures attributed to sinus epithelium and wall, structures which actually pertained to the surface epidermis?

INCIDENCE

Pilonidal sinuses are much more common than was supposed by earlier workers. This lesion is most common in early adult life—most reported cases occurring between the ages of 16 and 25 years. 18,19,30-32 It was once thought that the condition was rare in women 33; but, recent studies have shown this is not the case. 18,19,30-32 However, in most series the incidence in males is greater than that in females. 34-37

It was not until 1934³⁸ that a case was reported in a Negro. It had been believed that pilonidal sinus was a condition confined exclusively to the Caucasian race.³⁹ In 1935, Breidenbach and Wilson³⁰ reported 4 cases in Negroes in a series of 288 cases from Bellevue Hospital in New York City. By 1947, 21 cases had been reported.³⁶ It is interesting to note that in these reports the incidence is higher in females.³⁴

The occurrence of sacrococcygeal sinuses in identical twins has been reported by Goldberg and Bloomenthal,³³ Mechling,⁴⁰ and Fox.⁴¹ A familial incidence has been mentioned by Tendler^{36,42} and others.^{34,35}

ASSOCIATED ETIOLOGIC FACTORS

Many factors have been found to be more or less constant in cases of pilonidal sinus and have thus been designated as associated etiologic factors, or contributing causes. Two of these, hirsutism and poor local hygiene, were mentioned as early as 1880 by Hodges. Later papers emphasized these and other factors, such as trauma and obesity. Kallet, Kallet, in 1936, stressed adolescence as a factor associated with the development of pilonidal sinuses.

MATERIALS

A study series of 463 cases of pilonidal sinus was derived from the files of the Department of Pathology of the University of Michigan. Only the cases in certain years were used, but within those years the cases were unselected. All lesions were sacrococcygeal. The University Student Health Service supplied many cases; others were from University Hospital and various outside hospitals submitting surgical material for diagnosis. Pilonidal sinuses occurred in a ratio of approximately 1:500 specimens from other surgical procedures.

The few cases which had been diagnosed as congenital dermoid cysts, epidermoid cysts, and meningocele were excluded. The micro-

scopic sections from all cases were re-examined, and all gross material submitted with a clinical diagnosis of pilonidal sinus during a period of 7 months was examined carefully as it was sectioned.

RESULTS

Incidence. Since the cases were unselected and consecutive within the years used, clinical data were often incomplete. As to incidence in respect to sex, there were almost exactly twice as many males (304) as females (149) among those of a stated sex. This difference is probably unfairly weighted in favor of males because of male preponderance in the student body from which numerous cases were derived.

The age distribution by quinquennia, expressed in case units and also as a percentage of those of stated age, is shown in Table I. In

Age in years	No. of cases	Per cent of total	Age in years	No. of cases	Per cent of total
0-5	3	0.9	36-40	16	5.0
6–10	0	0.0	41-45	7	2.2
11-15	4	1.3	46-50	2	0.6
16-20	87	27.4	51-55	3	0.9
21-25	110	34.6	56-60	3	0.9
26-3 0	48	15.1	61-65	1	0.3
31-35	32	10.1	66-70	2	0.6
			Total	318*	100.0

TABLE I
Over-all Age Incidence

this series, 197 cases (62 per cent) occurred in the decade of 16 to 25 years of age. This may be over-weighted to some extent by the student cases which were included, but the figures are in general agreement with those of others and the age distribution must be considered significant.

In Table II, the distribution of both sex and age makes evident the somewhat earlier concentration in females. Cumulative percentages show that 38.8 per cent of the female cases occurred before age 21 as compared with 25.9 per cent in males before that age. The cumulative percentage for males is 60.3 before age 26; for females, 73.8. The earlier occurrence in females is believed to be significant and will receive further attention in the discussion of the pathogenesis.

Histopathologic Features. Microscopic examination revealed that the sinus opening on the cutaneous surface usually continued into a

^{* 68.68} per cent of total group of 463 cases.

deeper portion which, in many instances, showed cystic dilatation (Fig. 1). Often there was evidence of branching sinuses. A stratified squamous epithelial lining, of variable degrees of integrity, was found in 235 cases (51 per cent). Of the 463 cases, 334 (72 per cent) showed

Age groups	Males			Females					
	No. of cases	Per cent of total	Cumulative percentage	No. of cases	Per cent of total	Cumulative percentage			
0-5	2	1.0	1.0	ı	1.0	1.0			
6–10	0	0.0	1.0		0.0	1.0			
11-15	2	1.0	2.0	2	1.9	2.9			
16-20	50	23.9	25.9	37	35.9	38.8			
21-25	72	34.4	60.3	36	35.0	73.8			
2 6–30	30	14.3	74.6	14	13.5	87.3			
31-35	24	11.5	86. r	8	7.8	95.1			
36-40	13	6.2	92.3	3	2.9	98.0			
41-45	7	3-3	95.6	0	0.0	98.0			
46-50	2	1.0	96.6		0.0	98.0			
51-55	2	1.0	97.6	1	1.0	99.0			
5 6-6 0	3	1.4	99.0	0	0.0	99.0			
61-65	ı	0.5	99.5		0.0	99.0			
66-70	1	0.5	100.0	1	1.0	100.0			

TABLE II
Incidence in Relation to Sex and Age

Total number of known age and sex: 312.

Total number of males of known age: 209 (66.99 per cent of total).

Total number of females of known age: 103 (33.01 per cent of total).

hair shafts, either lying loose in the sinus, embedded in granulation tissue, or persisting deep in relatively mature scar tissue. In all instances, the hair stained as does "dead" hair.

A careful search was made for hair follicles which could be assigned to the wall of the sinus or cyst. In every instance in which follicles were present in the microscopic sections, it was evident that they belonged to the covering skin. The same was true of sebaceous glands, sweat glands, and arrectores pilorum muscles.

Infection and inflammation had a part in producing the general pathologic picture. Where there was no evidence of a squamous epithelial lining, the sinus was found to be lined by vascular pyogenic granulation tissue (Fig. 4). Cellular infiltrations consisted of polymorphonuclear leukocytes, lymphocytes, and plasma cells in varying proportions. Foreign body giant cells in association with dead hairs were a frequent finding (Fig. 2). In many cases there were large

mononuclear phagocytes containing blood pigment. Abscess formation deep in the tissues was present in 28.7 per cent of the cases.

DISCUSSION

From the review of the literature it appears that most writers have assumed that a pilonidal sinus results from a developmental lesion of some kind, and that the disturbance of growth has taken place at an early stage of fetal life. Among the reasons given for these assumptions are: (1) the presence in early fetal life of a connection of the skin with the neural canal; (2) the occasional presence of a deep sinus extending into the vertebral canal; (3) the occurrence of sacrococcygeal dimples; (4) the midline site of most sinuses, and (5) the presence of hair in the sinuses.

That a lesion such as a pilonidal sinus might well be associated with a deviation in anatomical structure is an obvious consideration. What is less obvious is that such a change, of which the results often are apparent only in adult life, must necessarily have occurred in the embryonic or fetal period. Hence it becomes necessary to examine more closely the arguments for the congenital origin of pilonidal sinuses.

- 1. Any anatomical structure or aggregation of tissues is significant only for a particular stage of development. The occurrence of a neurocutaneous communication in fetal life can be correlated intimately with skin lesions in postnatal life only by assuming that during all the changes which take place in the region of the neurenteric canal a part of this tract is relatively unaffected. Study of the stages of growth shows how completely earlier structures are changed into, or replaced by, entirely different ones. Moreover, the usual pilonidal sinus does not have tissue like that of the neurenteric canal nor does it approach the vertebral canal. There is little resemblance between spina bifida and pilonidal sinus. The former is present at birth. They occupy different sites, and the tissues involved are different.
- 2. Sinuses communicating with the spinal canal have been observed in the cervical and dorsal regions as well as in the lower portion of the vertebral column. Yet lesions comparable to the sacrococcygeal pilonidal sinus are not found in the superior regions.
- 3. Sacrococcygeal dimples are found in some infants, but no relationship to pilonidal sinuses has been observed. On the other hand, dimples are seldom found with sinuses³⁵ and at no stage of their development do pilonidal sinuses show morphologic similarities to dimples. A dimple with its smooth sloping edges and shallow base is quite different from the "pit" of a sinus, and it is always present at birth. The

sinus arises in tissues previously normal during childhood. However, there is no reason why a sinus might not develop within the area of a dimple, but evidence of a direct etiologic association is lacking.

- 4. The midline location of most sinuses has appeared to support the view of congenital origin on the ground of incomplete midline fusion of the lateral halves of the body. However, not all pilonidal sinuses occur in the midline 1,2,48; those which do not cannot be explained by this hypothesis.
- 5. The presence of hair in pilonidal sinuses and cyst is characteristic and is the particular feature which justifies the term in diagnosis. It was the presence of hair which attracted attention to the condition originally and which was responsible for the terminology applied to it. It is surprising to find that almost all writers subsequent to Anderson, Hodges, and Warren have assumed that the hairs arise locally, disregarding a possible extraneous origin. That the hairs do not arise from the sinus becomes quite clear when they are removed and examined. Hodges remarked: "The hairs . . . are always short, without bulbs." Sometimes a root is found, but then the root-end may be found to protrude from the mouth of the sinus. Warren, in 1867, even described the hairs as being "inverted."

Microscopically, hairs embedded deep in the sinuses are found to lie free in granulation tissue or scar tissue. They may be surrounded by foreign body giant cells. Hair follicles are never found in the walls of sinuses. These facts indicate that the hairs cannot come from within the sinuses, but are introduced from without. Such hairs might be broken off or shed locally, or they might be hairs from the back which had been shed and had then passed down between the buttocks.

The recognition of interdigital pilonidal sinuses as an occupational disease in barbers and the report of occasional examples since 1946 is obviously of prime importance in understanding the pathogenesis of the similar sacrococcygeal lesions.

In view of the objective evidence and other considerations as outlined, it seems that the theory of Patey and Scarff,² which explains pilonidal sinuses on an acquired, non-developmental basis, is better by far. In outlining a possible explanation for the occurrence of the lesions in both the postanal region and in the webs of the fingers, they divided its development into two phases: (1) an initial phase in which organisms are introduced into the tissues, there giving rise to the infection leading to sinus formation; and (2) the entrance of hairs into the sinus to produce the foreign body granulomatous reaction. Subsequent epithelization of the sinus from the surface may or may not take place, as is true, also, for the secondary abscesses and sinuses

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which may form. From experiments on the possible methods of the introduction of hair into a narrow orifice, Patey and Scarff² came to the conclusion that the hairs in pilonidal sinuses are sucked in by negative pressure and are not pushed in. They believed that in the webs of the fingers and near the postanal cleft, negative pressures are readily produced; in the webs of the fingers by the alternating tautening and loosening of the tissues during movements of the fingers, and near the postanal cleft by the alternating pressure and relaxation of the soft tissues against the coccyx and sacrum during sitting. This mechanical factor has been recognized by army physicians who have called pilonidal sinuses "jeep disease." The absence of negative pressures in other areas, even a short distance away from the postanal cleft, may explain why anal and ischiorectal abscesses show no tendency to become pilonidal.

The closeness of the buttocks, constant friction of skin and clothing, the accumulation of desquamated epithelium, hairs, and sebaceous material, and possible carelessness in personal hygiene make it easy to understand why the internatal region may be peculiarly vulnerable to the formation of crypts as a result of skin inflammation. Once formed, the negative pressure of the area may literally "aspirate" hairs into the pit.

Notwithstanding the experimental investigations by Patey and Scarff,2 the question may well be raised whether an inflammatory sinus or pit is necessary for the introduction of the first hair. Pilonidal sinuses develop where the skin is continuously moist and where there is no horny layer. It seems possible that a short, stiff hair might be forced through the macerated epidermis mechanically, to be followed by infection, inflammation, an actual sinus, and the eventual drawing in of enough hairs to constitute a small tuft. The use of dry paper in personal cleansing after defecation may be one method by which hairs can be forced through the epidermis. Dr. K. C. Samuel⁴⁹ of the Department of Pathology of Sawai Man Singh Medical College, Jaipur, India, has stated that in his surgical material sacrococcygeal pilonidal sinuses are very uncommon. Personal cleansing after defecation is by ablution and toilet paper is never used by the native population. While this suggestion is only hypothetic, there must be some explanation for the geographic difference in incidence. Dr. Samuel is certain that the reason is not to be found in any racial dissimilarity in hairiness.

That females reach puberty earlier than males and that the sex hormones play a large part in the regulation of the growth of body hair and secondary skin structures may be concerned with the earlier ap-

pearance of pilonidal sinuses in females. With the onset of puberty there may be a relatively greater concern over personal cleanliness and greater mechanical friction from clothing in women than in men.

Hirsutism is much more common in males. Males also lead more active lives, in occupation, sports, and military service. They are less concerned with bodily cleanliness and, in general, may be assumed to have a greater accumulation of shed hairs and other detritus in the postanal region. These seem to be important reasons for the greater incidence of pilonidal sinuses in males.

It is less easy to explain the concentration of pilonidal sinuses in the period between the ages of 16 and 35. It seems not entirely adequate to note that these are the years of the rapid development of hair along with increased sebaceous gland activity, and also the years of increased physical activity. It may well be that those who, by personal habits and by character of hair and of epidermis, are proper subjects for acquiring a pilonidal sinus will have done so by age 35.

SUMMARY AND CONCLUSIONS

Study of a series of 463 cases of pilonidal sinus established that, as compared to other surgical specimens, the incidence was roughly 1:500 in the material examined. The lesion is more common in males than in females and usually becomes apparent during the second or third decade. The frequency distribution as to age and sex shows that pilonidal sinuses tend to appear about 5 years earlier in females than in males.

The pathologic picture is that of an acute or chronic inflammatory dermal sinus which contains dead hairs in about three fourths of the cases. Hair follicles are never found in the walls of these sinuses.

A review of the literature and correlation of the objective information with the various theories of pathogenesis has led to the rejection of the commonly accepted developmental or congenital theory in favor of the opinion that sacrococcygeal pilonidal sinuses and abscesses are acquired. The recognition of similar pilonidal sinuses of the interdigital web as an occupational disease of barbers provides strong evidence for a similar mode of origin for the more common sacrococcygeal lesions.

ADDENDUM

The careful study of the interdigital sinuses of barbers' hands by Currie, Gibson, and Goodall ⁵⁰ was noted after this paper had been accepted. These authors reported 11 new cases of this occupational disease, bringing the total described in the literature to 29. They emphasized the importance of short, sharp hairs in causation and of personal hygiene in prevention. They prefer to restrict "pilonidal sinuses" to the postanal or perineal lesions and to designate the condition described by them as

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"interdigital sinuses of barbers' hands." In the same journal, Hueston⁵¹ discussed the pathogenesis of pilonidal sinuses of the sacrococcygeal region and concluded that they are acquired lesions of foreign-body type due to the penetration of short stiff hairs.

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LEGENDS FOR FIGURES

- Fig. 1. The dilated fundic portion of this epithelium-lined tract is nearly filled with dead hairs. The epithelium is intact. No hair follicles or other accessory epidermal structures are present. Hemalum and eosin stain. × 33.
- FIG. 2. About twenty-five dead hairs lie nearly parallel in this pilonidal sinus. The epithelium produces abundant keratohyalin which is desquamating into the lumen. There are no hair follicles. Hemalum and eosin stain. X 110.
- FIG. 3. Early foreign body giant cell reaction around dead hair shafts in the granulation tissue lining a pilonidal sinus. Hemalum and eosin stain. X 110.
- Fig. 4. Dead hair shafts surrounded by newly formed connective tissue in the lumen and wall of a pilonidal sinus. Hemalum and eosin stain. X 110.

