

Research Article

Unusual Sites of Pilonidal Sinus, What is The Difference?

Elsaady A*

Departement of General Surgery, Kafr Elshikh General Hospital, Egypt

***Corresponding author:** Ahmed Elsaady, Departement of General Surgery, Kafr Elshikh General Hospital, Egypt**Received:** February 22, 2019; **Accepted:** March 28, 2019; **Published:** April 04, 2019**Abstract**

Pilonidal sinus can be developed in unusual areas. This is a prospective study, done to assess patient characteristics, risk factors & effective treatment modalities of the disease in such sites & compare to that developed in the sacrococcygeal area. Patients presented with pilonidal sinus within five years were involved in the study & divided into; group (1) of unusual site & group (2) with sacrococcygeal sinus. The study evaluated age, gender, hirsutism, deep cleft, lack of hygiene, obesity, frictional impact & effective management modalities (conservative & or surgical) & compare both groups. Two hundred and fifty one cases were presented, with forty nine included in group (1) & two hundred and two cases included in group (2). Varieties of group (1) included umbilical, interdigital, axillary, inter-mammary, gluteal & inetrgluteal with anal extension. Risk factors as hirsute, deep cleft, lack of personal hygiene & frictional impact are significant in this group. Conservative measures had significant role in this group, with the operative interference was significantly more demanding. The study concluded that; pilonidal disease can be developed in unusual sites as long as the risk factors (as loose hair, deep cleft, & frictional impact) are present. Some aspects of the disease in these areas differ from that developed in intergluteal one, with the conservative measures is quite effective to start with. Surgical management varies from simple excision to wide excision & refashioning of new cleft. This may be more demanding and requires considerable cosmetic and functional outcome prognostic issues.

Keywords: Pilonidal sinus in unusual areas; Rare sites of pilonidal disease; Umbilical pilonidal sinus; Web space pilonidal disease

Introduction

Pilonidal disease is a relatively common disease [1], that was originally reported by Anderson as a hair detected in a sacrococcygeal ulcer in 1847 [2]. It was firstly defined as pilonidal sinus by Hodges in 1880 [3]. Although there was a great debate in the etiology of the disease either congenital or acquired, but most authors now believe that the disease is acquired [4]. The disease occurs mostly in the sacrococcygeal area, but several unusual occasional sites have been reported in literatures (including umbilicus, forehead, scalp, clitoris, inter-digital area, penis, abdomen, neck and axillae [5]. This study is a prospective one, that aims at demonstrating the characteristics of the disease developed in unusual sites in comparison to that developed in sacrococcygeal area.

Materials & Methods

Over five years, all patients presented with pilonidal sinus in the sacrococcygeal area as well as unusual sites (from 3/2015 till 4/2018) were involved in the study. Patients were divided into two groups; group (1) patients with the sinus at unusual sites, & group (2) with sacrococcygeal sinus. Diagnosis was made by the presence of history of repeated inflammations and discharges from one or more openings with demonstration of broken hairs seen exuding or present within the sinus, or histological evidence of the disease in some cases of unusual sites. Each group was studied to assess patient characteristics, risk factors accused for development of the disease, and management modalities. The study assessed the presence of; invaders (hair), force of penetration and & vulnerability of skin for hair penetration in a

deep cleft (anatomical predisposition) in every case. The management entails three pivots; (1) Hair removal by regular shaving or depilatory agents or even laser ablation {in two cases}. Also, good personal hygiene, local cleanliness & sometimes weight reduction were advised. This constitutes the conservative management which was tried as a routine for at least three months, (2) Surgical excision of the sinus & refashioning the anatomical predisposition (if available) if the conservative measures failed. (3) Minimizing the force of penetration, this may entail changing the career like barbers, drivers, or some changes in behavior and dressing with get off tight cloths, and weight reduction for obese patients. Some operative issues such as the rationale of the procedure, operative time, the feasibility & need of consultation, the duration of healing, as well as complications and recurrence rate were also involved in the study. The patients requests for good cosmeses & or functional anatomy were also considered. The two groups were compared in the previous items to demonstrate if there real difference between them or not. Also, assessment of importance of the risk factors as well as efficacy of the management rationale was done. Patients were followed up for a minimum of eight months. We exclude cases that dropped in follow up.

Results

Two hundred and fifty one cases were presented in this study. Group (1) included forty nine cases representing nearly 20% of all cases, while group (2) included two hundred and two cases (~80% of all cases). The umbilical pilonidal sinus was the commonest form of group (1), affecting thirty three patients (~67%). This variant presented in the study in different clinical varieties, including abscess,



Figure 1: Chronic discharging sinus or eczema from chronic discharge.



Figure 2: The patient's characteristics and details.



Figure 3: Relatively wider shallower inter-mammary groove.

or chronic discharging sinus or eczema from chronic discharge as in Figure 1. Seven cases (~14%) of group a presented with axillary pilonidal sinus. Two cases (~4) presented with pilonidal fistula. This entails anal extension of the pilonidal disease (proved by contrast study). Other two cases presented with web space lesion. The remaining cases two case developed in the inter-mammary, and two cases in gluteal region so far from the sacrococcygeal area. Figure 2 show some of these cases.

The patient's characteristics and details in group (1) included that the mean age of this group is 25 years, with males affected in 65% of cases. Excessive & or dense hair found in 94% of cases, while lack of personal hygiene reported in 70% of cases. Anatomical predisposition of the sites affected with the presence of a deep cleft presented in 94% of cases. The availability of the force of penetration & vulnerability of skin for penetration were manifested in 94%. The average duration of complain was 1.7 months. Conservative measures were successful in thirty one (~63%) cases. Surgical treatment varies from simple excision with conservative measures in seven (~39%) cases, excision and refashioning of the cleft in eleven (~61%) cases. The average operative time was nearly 45 minutes. General surgeons operated fourteen (~78%) cases with three cases done by general surgeon after consultation (16%) & one did by plastic surgeon (~6%). The average healing period was about twenty-six days. Patients asked for good cosmeses & or good functional anatomy in eleven (~61%) patients. Postoperative complications occur in two (~11%) cases, all of them are minor complications (mild seroma) with no recurrence reported.

On the other hand, details of patients developing the disease in the usual sacrococcygeal area entailed that the mean age of this group is 21 years, with males affected in 85 %of cases. 94% of cases

were hirsute, while lack of personal hygiene reported in 49% of cases, and deep cleft manifested in 98% of them. The force of skin penetration was reported in 80%. The average duration of complain was seven months. Successful conservative measures were achieved only in thirty-nine (~19%) cases. Surgical treatment which consists of Karydakis technique has an average operative time of about 55 minutes & healing period of about 19 days. Complications occurred in thirty two (~16%) cases including; seroma in eighteen cases (11%), wound dehiscence in twenty seven cases (13%), hypertrophic scar in four cases(2.4%) & painful scar in ten cases (6.1%) and recurrence in eight cases (~5%).

Comparison of both groups (Table 1) demonstrates that there is no statistically significant differences in the age affected, obesity as risk factor, as well as operative time, complications and the duration of healing. The duration of complains is insignificant, but shorter in unusual sites, which may be attributed to their apparent sites. But on the other hand a significant difference is well evident in some characters between both groups. The presence of the risk factors such as hirsute, deep cleft & lack of personal hygiene are significantly well evident in unusual sites (group 1) which denotes their importance in the development of the disease in such sites. Females are significantly affected in group 1 than group 2. Also, conservative measure succeeds significantly in group 1 than group 2. A special demand was needed in operative interference in group1, where we found the need for consultation of others (like plastic or colorectal surgeons) is significant in this group, and also the patient's request for good functional or & cosmetic outcome is significant & prominent in this group. This can be attributed to many factors such as more female affection, prominent sites & relation to the occupational needs & skills as in barber. The rate of recurrence is significantly lower in unusual sites. The

Table 1: Comparative studies between patients develop pilonidal disease at unusual sites group (1) and those with sacro-coccygeal disease group (2).

Item of study	Group (1) (Unusual sites)	Group (2) (Intergluteal area)
Number of cases	49 (~20%)	202 (80%)
Age;		
Mean;	25.5	21
Standard Deviation;	4.71	4.2
Levene`s test for equality of variances	0.214	insignificant
Gender;	Male; Female 31; 18 Females ~37%	Male; Female 170; 32 Females ~16%
Mean	1.64	1.85
Standard Deviation;	0.492	0.355
Levene`s test for equality of variances	0.002	Significant
Lack of personal hygiene;	37/49	99/202
Mean	0.76	0.49
Standard Deviation;	0.4372	0.5045
Levene`s test for equality of variances	.000	significant
*Anatomical predisposition (cleft)	46/49 (94%)	197 /202 (98%)
Mean	0.94	0.76
Standard Deviation	0.2425	0.4287
Levene`s test for equality of variances	.000	significant
Obesity & or manifest weight gain	27/49	109/202
Mean	0.55	0.54
Standard Deviation;	0.4925	0.5025
Levene`s test for equality of variances	0.083	insignificant
Duration of complain	1.7 months	3 months
Mean	10.8	6.8
Standard Deviation	3.339	2.771
Levene`s test for equality of variances	0.264	insignificant
Successful conservative treatment	31/49	39/202
Mean	0.23	0.03
Standard Deviation;	0.4372	0.1889
Levene`s test for equality of variances	.000	significant
Operative issues		
Operative time (in minutes)		
Mean	46.4	54.7
Standard Deviation;	20.74	20.72
Levene`s test for equality of variances	0.955	insignificant
Rate of Complication	2/18 (11%)	32/163 (16%)
Mean	0.15	0.18
Standard Deviation;	0.3132	0.4372
Levene`s test for equality of variances	0.155	insignificant
Rate of Recurrence;	0/18 (0%)	8/163 (5%)
Mean	.00	0.05
Standard Deviation;	.000	0.2333
Levene`s test for equality of variances	0.001	significant
Consultation;	3 (~16%)	0

Mean	0.23	.00
Standard Deviation;	0.4385	
	.000	significant
Request of good Cosmoeses & or good functional anatomy		
Mean	0.46	0.0943
Standard Deviation;	0.5187	0.2951
Levene`s test for equality of variances	.000	significant
Healing period (in days)		
Mean	17	20
Standard Deviation;	24.77	8.714
Levene`s test for equality of variances	0.955	insignificant

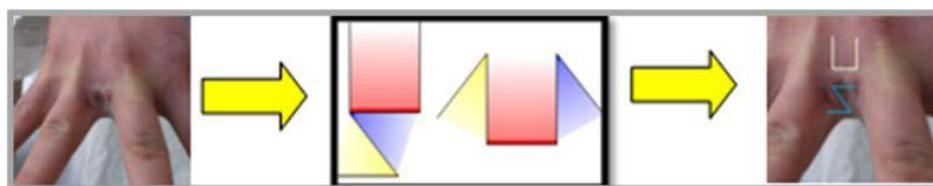


Figure 4: Square flap with Z-plasty.

management approach targeting the risk factors used in this study had few complications & no recurrence reported in group (1) and eight cases (5.%) of group (2).

Discussion

Pilonidal sinus is a relatively common disease [1]. The disease represents chronic foreign body reaction for hair & is characterized by a granulomatous reaction to a hair shaft penetrating the epidermis from the cutaneous surface [4]. Patey and Scarff (in 1940-1950) suggested hair movement from the surrounding skin under frictional impact, & may puncture the skin initiating the process [7]. It is speculated that a cleft creates a suction that draws hair into the groove which pits on movement and friction [1]. Accordingly, the development of the disease is very suggestible by the concomitant availability of; invader (loose hair), force causing hair penetration & skin vulnerability for penetration in the depth of a cleft [7]. Accordingly, once the three factors are provided at any sites (whatever it is unusual) the disease may be developed [5]. On practice these factors entails some predisposing factors that increase the risk for development of the disease. Invader tends to be more in hirsute, male, lack of personal hygiene, & some occupations such as barber, sheep shearers, & milker [8], while some anatomical predispositions may pave the way for the disease development such as intergluteal cleft, axillae, web space, umbilicus [9]. Obesity & or recent evident weight gain usually make such cleft deeper providing better trapping field [10]. On the other hand, some factors such as prolonged sitting in moving vehicles & jeep drivers [11], movement and frictions of fingers in barbers and hair dressers [12], & frequent movement of upper limb [13], especially the dominant one increases the force of penetration and add greatly to the risk of development of the disease in sacrococcygeal, web space and axillae respectively once other risk factors present. In this study, cases of unusual sites (group 1) represent nearly 20% of all cases which is a large number compared to the usual description in literature for these unusual sites as rare presentations

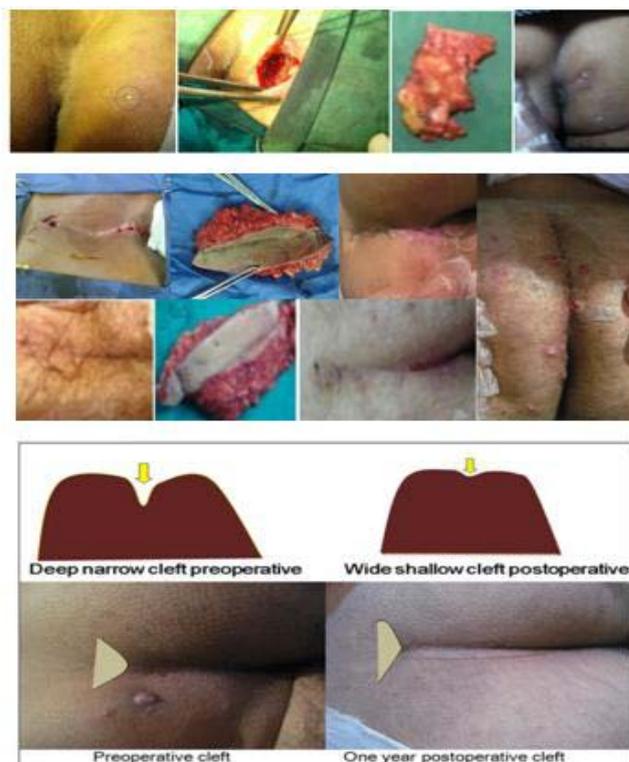


Figure 5-7: The gluteal case simple excision of the sinus.

[5]. This may demonstrate actual alarming increase in the incidence, but still need more and more studies. Such surprising incidence may be attributed to the higher incidence of risk factors nowadays such as obesity [14], socioeconomic issues with long periods out-home life that may be associated with lack of personal hygiene & tight cloths [15]. The study demonstrates the great importance of the presence

of hair, deep cleft & frictional impact force in the development of the disease especially in unusual sites. All umbilical cases have deep umbilicus, and mostly obese, & hairy male. Case of intermammary affection had a cleft or groove between pendulous breasts, this in the presence of bra and movement provided the force of penetration of loose hair coming from being hirsute. Cases of web space affection belongs to barbers. His occupation gave the source of loose hair and frequent movement & friction of fingers providing the power of penetration to the anatomical cleft already present [16]. The cases of axillary affections are hirsute, with the development of the disease in the dominant side, which may be attributed to more movement than the other sides. Friction (abduction-adduction) [17], suction, massage, shaving, pounding, minor infection and maceration are assorted mechanisms which play a part in development of the disease in axillae [18]. The cases affecting the gluteal region although there was no cleft, but obesity, tight rough cloth, being driver & hairy may provide considerable risk factors for the development of the disease in one case but the other was a female, non obese patient but hirsute.

The natural history of the disease in untreated patients suggests that, in some cases the disease disappears with time [5]. Accordingly, conservative measures were applied initially for all patients to relief the acute inflammatory states and to make patients stick to regular removal of hair & hygienic measures for minimizing the risk of recurrence [19] and sometimes hoping for its sunset [20]. This study reported an effective role in conservative management in group 1 (unusual sites) compared to the usual sacrococcygeal disease. Umbilical pilonidal sinus responds well to conservative measure [20] and this study demonstrated success in 63% of cases. So conservative measures should be the first line of treatment in all cases of unusual sites & may be tried in the sacrococcygeal group specially in older patients.

On management of the disease the three factors should be targeted to eliminate or minimize the risk of recurrence as much as possible, which entail abolishing or minimizing; (a) the invaders by removal of hair, (b) the cleft by excision and refashioning & (c) the force for penetration by close sticking to conservative measures. So the management should be based on three pivots;

Hair removal; by regular shaving or using depilatory agents or even laser ablation {in two cases of group (2); hirsute as recommended in some literature [21]. Also we insist on good personal hygiene and local cleanliness.

Refashioning the anatomical predisposition if available after excision of the sinus. For umbilical pilonidal diseases sinusectomy [22], omphalectomy without reconstruction [23] or omphalectomy with reconstruction of new umbilicus [15] were described in literature. The new umbilicus should be cosmetically accepted and less risky for recurrence [24]. Here we did omphalectomy with reconstruction of new umbilicus. The excision occasionally may extend more than the umbilicus (in two cases) according to the extent of indurations. We insist that the new umbilicus should be shallow, wide, looking downward rather than deep, narrow & looking upward to minimize the risk of trapping. This is done by remove a part of subcutaneous fat in area prepared to be the new umbilicus then pull & fix the dermis above to the rectus sheath in a line rather than a point by transverse mattress suture or a loose purse string without tightening so that

the new umbilicus become fixed in a ring. Such umbilicus provides cosmetically accepted one with minimal risk for recurrence.

As regard to the inter-mammary disease simple excision will lead to more encroachment of breasts and subsequently more grooving, also closure in the ellipse in the midline carries high risk of keloid development such factors made a plastic surgeon to operate where he excised along the black markers and extended along red lines to allow closure along the radius extent of the inferior breast folds, creating relatively wider shallower inter-mammary groove as in Figure 3, minimize the risk of recurrence.

On web space disease, simple elliptical incision will end in more narrow web space, which entails higher recurrence & limited movement of the fingers which does affect occupational skills [16]. Here the sinus is excised within Z-plasty & the refashioning achieved by advanced square flap; the so-called square flap with Z-plasty as in Figure 4. This technique gave a wide area for the web reconstruction, which allows free movement of fingers & wide shallow cleft. For the gluteal case simple excision of the sinus as in Figures 5-7 were done with restricts measures about hair removal. Also, in axillary pilonidal disease simple excision were done with strict conservative measures. This was described to be sufficient in literature [25]. The cases with anal extension; elliptical excision with fistulotomy at the distal anal part. The internal opening was low at 6th. O'clock. Marsupialization was done in the middle coccygeal part, while the proximal sacral part was closed in layers. Three in one technique, which entail fistulotomy, marsupialization and closure in layers, hoping for more rapid healing and less pain than to leave the total wound opened.

For sacrococcygeal disease (group 2) Karydakakis technique was used to allow proper excision of the sinus & flattening the previous deep intergluteal cleft through a lateral incision [26].

Minimizing the force for penetration, this actually were difficult practically, because its measures may entail changing the career like barbers, drivers, or some changes in behavior and cloth like get off tight cloths, in addition to weight reduction for obese patients.

This study demonstrated that the management approach targeting the risk factors of development of the disease; hair, cleft, & vulnerability for skin penetration is very effective tool in the treatment of pilonidal disease anywhere.

Conclusion

Pilonidal disease can be developed in unusual sites away from the intergluteal groove, with incidence may be more than before. This study demonstrated significant differences in some aspects of the disease in these areas compared to that developed in sacrococcygeal one. The presence of risk factors is essential in the development of the disease in such unusual sites. These factors include the presence of hair, deep cleft, as well as frictional impact. Management should target the risk factors to be eliminated or decreased as much as possible. Conservative measures are quite effective to start with in these sites. Surgical management varies from simple excision of the sinus to wide excision of the sinus & refashioning of new cleft. This may be more demanding and requires considerable cosmetic and functional outcome prognostic issues as it may affect the career of the patient. Although this study was small, but it necessitates to spot lights on this subject and do further assessment & studies.

References

1. Jeffrey A. Sternberg. The management of pilonidal disease. In: Current Surgical Therapy. Cameron JL, et al., 12th edition, Philadelphia, Saunders Elsevier. 2017; 387-390.
2. Anderson AW. Boston. Med Surg J. 1847; 36: 74.
3. Hodges RM. Boston. Med Surg J. 1880; 103: 485.
4. Kelli M, Dunn B, Rothenberger DA. Pilonidal disease. Colon, Rectum, and Anus. Schwartz's Principles of Surgery F. Charles Brunicaudi. 1880; 29: 1233-1234.
5. Sion-Vardy N, Osyntsov L, Cagnano E, Osyntsov A, Vardy D, Benharroch D. Unexpected location of pilonidal sinuses. Clin Exp Dermatol. 2009; 34: e599-601.
6. Keighley M. Anorectal Disorders. Fischer's Mastery of surgery, Josef E. Fischer, William V. McDennott, Christian R. Holmes .sixth edition. 2012; 171; 1811-1814.
7. Hull TL, Wu J. Pilonidal disease. Surg Clin North Am. 2002; 82: 1169-1185.
8. da Silva JH. Pilonidal cyst. Dis Colon Rectum. 2000; 43: 1146.
9. Karakayali F, Karagulle E, Karabulut Z, Oksuz E, Moray G, Haberal M. Unroofing and marsupialization vs. rhomboid excision and Limberg flap in pilonidal disease: a prospective, randomized, clinical trial, Dis Colon Rectum. 2009; 52: 496-502.
10. Blereau RP. Pilonidal Cyst of the Umbilicus. Cancer Network September. 2005; 14.
11. Jan Rakinic. Sacrococcygeal pilonidal sinus. In Current Surgical Therapy John L. Cameron. Mosby Ninth Edition. 2008; 70: 302-305.
12. Al-Khamis A, McCallum I, King PM, Bruce J. Healing by primary *versus* secondary intention after surgical treatment for pilonidal sinus. Cochrane Database of Syst Rev. 2010.
13. Oryu F, Minagawa H, Chiba N. Three cases of a pilonidal sinus of the axilla. Clin Dermatol. 1992; 34: 1631-1634.
14. Eryilmaz R, Sahin M, Okan I, Alimoglu O, Somay A. Umbilical pilonidal sinus disease: predisposing factors and treatment. World J Surg. 2005; 29: 1158-1160.
15. Elsaady A, Aboelsaad M, Elbeheery M. Umbilical Pilonidal Disease; Predisposing Factors & Best Modality of Management. Journal of surgery. 2018; 6.
16. Stern PJ, Charles A. Goldfarb. Interdigital Pilonidal Sinus. N Engl J Med. 2004; 350: e10.
17. Sengul I, Sengul D, Mocan G. Axillary pilonidal sinus: A case report. N Am J Med Sci. 2009; 1: 316-318.
18. Imamura T. A case with a pilonidal sinus of the axilla (abstract). Jpn J Dermatol. 1993; 103: 1467.
19. Aydede H, Erhan Y, Sakarya A, Kumkumoglu Y. Comparison of three methods in surgical treatment of pilonidal disease. ANZ J Surg. 2001; 71: 362.
20. Coşkun A, Buluş H, Akıncı OF, Özgönül A. Etiological Factors in Umbilical Pilonidal Sinus Indian J Surg. 2011; 73: 54-57.
21. Odili J, Galt D. Laser depilation of the natal cleft-an aid to healing the pilonidal sinus. Ann R Coll Surg Engl. 2002; 84: 29.
22. Naraynsingh V, Hariharan S, Dan D. Umbilical Pilonidal Sinus: A New Treatment Technique of Sinus Excision with Umbilical Preservation Dermatol Surg Published by Wiley Periodicals. 2009; 35: 1155-1156.
23. Akkapulua N, Tanrikulua Y. Umbilical Pilonidal Sinus: A Case Report. J Med Cases. 2011; 2: 272-274.
24. Akba H, Guneren E, Eroglu L, Uysal OA. Natural-looking umbilicus as an important part of abdominoplasty. Aesthet Plast Surg. 2003; 27: 139-142.
25. Ohtsuka H, Arashiro K, Watanabe T. Pilonidal sinus of the axilla: report of five patients and review of the literature. Ann Plast Surg. 1994; 33: 322-325.
26. Karidakis GE. Easy and Successfull treatment of pilonidal sinus after explanation of its causative process. Aust NZJ Surg. 1992; 62: 385-389.