original article

Pilonidal sinus disease recurrence at a tertiary care center in Riyadh

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BACKGROUND: Pilonidal sinus disease (PSD) is a chronic inflammatory disease of the sacrococcygeal area. Pilonidal sinus recurrence is a matter of concern to both patients and healthcare providers.

OBJECTIVES: Estimate the rate of PSD recurrence in our center and identify any risk factors contributing to disease recurrence.

DESIGN: Retrospective.

SETTING: Tertiary care center in Riyadh.

PATIENTS AND METHODS: All patients who underwent surgery for primary or recurrent pilonidal sinus between 1 January 2016 to 30 June 2019 were included to allow for at least 1-year of follow-up at the time of data collection.

MAIN OUTCOME MEASURES: Recurrence rate of PSD and risk factors for recurrence.

SAMPLE SIZE AND CHARACTERISTICS: 369 patients: 329 males (89.2%) and 40 (10.8%) females. Median (interquartile range) age was 21 (18-26) years.

RESULTS: Of the 369 included patients, recurrence was identified in 84 (22.8%) cases [95% confidence interval (CI) 18.6-27.4], and the mean timing of recurrence was 1.8 (1.6) years after the primary surgery. In a multivariate logistic regression analysis, increased age and post-operative seroma fluid discharge were independent risk factors for recurrence. In contrast, preoperative antibiotic prophylaxis and postoperative hair removal were effective in reducing recurrence. Type of surgery closure had no effect on recurrence, yet primary closure was associated with early onset of recurrence compared to secondary closure (P=.02).

CONCLUSION: Our findings on the factors associated with recurrence of PSD are consistent with many reports in the literature. Reported prevalence estimates vary widely.

LIMITATION: Single center, small sample size, retrospective.

CONFLICT OF INTEREST: None.

Pilonidal sinus disease (PSD) is a chronic inflammatory disease of the sacrococcygeal area. PSD commonly affects young males more than females and has an estimated incidence of 26 per 100 000 people.[1] It was initially thought to have a congenital origin, but is now thought to be an acquired disease related to the accumulation of hair in the natal cleft causing a foreign body reaction and inflammation.[2,3] The main predisposing factors for PSD include obesity, prolonged sitting, family history, chronic trauma, and irritation such as driving, type of work, poor local hygiene, taking a bath two times or fewer a week, and hirsutism.[4,5] Pilonidal sinus recurrence is a matter of concern to both patients and healthcare providers. Management of pilonidal sinus varies from wide excision to antibiotics alone. Surgical excision of PSD is the mainstay of treatment, but recurrence remains common. The reported PSD recurrence rates vary from one study to another and there is a limited understanding of predictors of recurrence.[6-8]

While some peer-reviewed articles have sought to determine risk factors for pilonidal sinus recurrence, none have shown decisive evidence. Thus, due to the shortfall of evidence-based research on this topic, the risk factors associated with pilonidal sinus recurrence are unclear. The aim of this study was to estimate the rate of PSD recurrence in our center and to identify any risk factor that contributed to disease recurrence.

PATIENTS AND METHODS

This single-center retrospective study was conducted at King Abdulaziz Medical City, Riyadh, Saudi Arabia. All patients who underwent primary or recurrent surgery for pilonidal sinus disease between 1 January 2016 to 30 June 2019 were included to allow for at least 1-year of follow-up at the time of data collection. All patient and disease characteristics were obtained from the electronic health record system, which provided the time of the primary surgery. Only the first episode of recurrence was included in the analysis. In addition, phone interviews of all patients were conducted to complement the patient information. Patients who were diagnosed with pilonidal sinus disease and did not undergo surgery or missed the scheduled follow-ups were excluded from the study. The study was approved by the Institutional Review Board at King Abdullah International Medical Research Center. Every patient contacted by phone agreed to take part in the interview.

Pilonidal sinus disease recurrence following surgical excision was set as the primary outcome of interest. Patients were diagnosed as having disease recurrence through outpatient surgery clinic records if they met at least one hard or two soft recurrence criteria illustrated by Doll (**Table 1**).[9] The following patient, disease, and surgery characteristics were examined as probable risk factors for PSD recurrence: gender, age, body mass index (BMI), smoking, family history of PSD, sitting habit, type of occupation, type of pilonidal sinus, type of primary surgery, preoperative antibiotic prophylaxis, history of incision and drainage, post-operative wound infection, post-operative seroma fluid, post-operative wound dehiscence, post-operative sitz bath, and post-operative hair removal. History of prolonged sitting was defined as sitting for more than 6 hours a day. Post-operative seroma fluid was defined as any fluid discharge noticed during wound dressing change visits not related to wound infection. Pilonidal sinus type was categorized into two groups: acute and chronic.

The surgical technique used in our hospital was either simple primary midline closure or lay open. Primary midline closure is an excision of the sinus with a direct midline closure of the wound by stitches while lay open is a wide excision of the sinus leaving the wound open to heal by secondary intention. Type of surgery was classified into two mutually exclusive groups: primary closure or secondary closure as described in the primary surgery note. Postoperatively, suction drains following excision with primary closure were not used.

The use of antibiotics for PSD has been evaluated only in preoperative prophylaxis, defined as the administration of a single prophylactic dose of intravenous antibiotic in the operating room prior to surgical incision. In this study, the choice for prophylactic antibiotics was cefazolin for all the patients.

Statistical analysis

Continuous variables are expressed as median and interquartile range (25th to 75th percentile) while categorical variables are reported as frequencies and percentages. Univariate analysis including Pearson's

Table 1. Criteria for recurrence assessment after complete wound healing of pilonidal sinus surgery.

Hard criteria	Soft criteria
Diagnosis by a physician	Redness
Incision (reintervention)	Swelling
Excision (reoperation)	Pain
Formation of a new sinus	Discharge
Presence of hair in a sinus opening	
Discharge of pus	

chi-square and Wilcoxon rank sum test with continuity correction were used to compare categorical and continuous variables, respectively. A multivariate logistic regression model was used to detect possible risk factors that might independently affect recurrence. A univariate regression model was used to assess the association between the type of surgery closure and time to recurrence. These statistical analyses were computed using IBM SPSS, version 24 (IBM, Armonk, NY). A result was considered significant at *P*=.05.

RESULTS

Of the included patients, 329 (89.2%) were male and 40 (10.8%) were females (**Table 2**). The median (IQR) age was 21 (18-26). The median BMI (IQR) was 28.7 (25.3-33.7). Most patients were non-smokers and had no comorbidities and not different between the groups; however, if comorbidity was present, then anorectal conditions were the most common comorbidities reported 17 (4.6%) followed by dyslipidemia 14 (3.8%), diabetes mellitus in 11 (3%), and hypertension in 9 (2.4%). Nearly, half of the sample were students (49.9%). More than one-third had a positive family history of PSD. About two-thirds of the patients reported a history of prolonged sitting.

Approximately 90% of our patients had chronic PSD, and only 39 patients (10.6%) had acute PSD with abscess (**Table 3**). We found that 207 patients (56.4%) underwent primary closure and 161 (43.6%) patients had secondary closure (healing by secondary intention); 290 (78.6%) took preoperative antibiotic prophylaxis. Regarding postoperative wound complications, 29 (7.9%) patients had wound infection, 185 (50.1%) experienced wound fluid discharge, and only six patients (1.6%) had wound dehiscence. Nearly half of the patients had hair regularly removed after surgery n=190 (51.5%) and 171 took a sitz bath postoperatively (46.3%).

Recurrence was identified in 84 (22.8%) cases (95% confidence interval (CI) 18.6-27.4). Using univariate analysis, the factors associated with PSD recurrence were age, BMI, hair removal postoperatively, post-operative seroma fluid discharge, and preoperative antibiotic prophylaxis. In a multivariate regression model factors associated with PSD recurrence included advanced age and postoperative seroma fluid discharge. On the other hand, the risk of PSD recurrence would likely decrease for those who received antibiotic prophylaxis preoperatively and those who underwent postoperative hair removal. In contrast, prolonged sitting did not have a significant effect on PNS recurrence (**Table 4**).

Although the type of surgery closure was not associated with higher recurrence rate (*P*=.50), the early onset of PSD recurrence was significantly more associated with patients who received excision with primary closure, and the likelihood ratio was 1.4 times higher than secondary closure (OR=1.45; CI=1.04 – 2.03; *P*=.02). The mean recurrence time of patients who received excision with primary closure was 1.4 (1.3) years, and the mean recurrence time of patients who received excision with secondary closure was 2.3 (1.8) years. The overall mean recurrence time was 1.8 (1.6) years.

DISCUSSION

Patients with pilonidal disease suffer considerable morbidity related to disease recurrence. Our results over a minimum of 1-year follow-up show a recurrence rate of 22.8% in patients treated surgically for pilonidal sinus.

Table 2. Patient characteristics (n=369).

Factors	Recurrent (n=84, 22.8%)	Non-recurrent (n=285, 77.2%)	P value
Age (years)	20 (19-27)	22 (17-23)	**<.001**
Gender (male/female)	76/8	253/32	.65
BMI (kg/m^2)	28.4 (25.1-33.5)	28.8 (25.5-33.7)	**<.001**
Smoking history			
Yes	21 (25)	84 (29.5)	.42
No	63 (75)	201 (70.5)	
Family history of pilonidal sinus disease			
Yes	37 (44)	94 (33)	.06
No	47 (56)	191 (67)	
History of prolonged sitting			
Yes	58 (69)	186 (65.3)	
No	15 (17.9)	30 (10.5)	.242
Unknown	11 (13.1)	69 (24.2)	
Occupation			
Office worker	8 (9.5)	28 (9.8)	
Healthcare worker	1 (1.2)	10 (3.5)	
Military	8 (9.5)	37 (13)	.36
Student	49 (58.3)	135 (47.4)	
Unemployed	7 (8.3)	18 (6.3)	
Unknown	11 (13.1)	57 (20)	

Data are n (%) or median (interquartile range).

However, there is considerable variation in the recurrence rates reported in the literature ranging from 0% to 100%.[10,11] One meta-analysis suggest that the variance in recurrence is due to different follow-up periods and that applying a recurrence without identifying the follow-up period after surgery may lead to a bias factor of up to 18 and above.[10] Recently, Doll et al showed that PSD recurrence is also influenced by geography after comparing recurrence rates of specific surgical approaches between four geographical regions.[12]

Table 3. Disease characteristics and surgery-related characteristics (n=369).

Factors	Recurrent n=84 (22.8%)	Non-recurrent n=285 (77.2%)	*P* value
Type of pilonidal sinus			
Acute with abscess	5 (6)	36 (12.6)	.08
Chronic	79 (94)	259 (87.4)	
Type of surgery			
Primary closure	50 (59.5)	158 (55.4)	.50
Secondary closure	34 (40.5)	127 (44.6)	
Antibiotic prophylaxis			
No	27 (32.1)	52 (18.2)	.006
Yes	57 (67.9)	233 (81.8)	
History of incision and drainage			
No	74 (88.1)	233 (81.8)	.17
Yes	10 (11.9)	52 (18.2)	
Postoperative wound infection			
No	78 (92.9)	262 (91.9)	.78
Yes	6 (7.1)	23 (8.1)	
Postoperative wound dehiscence, n (%)			
No	83 (98.8)	280 (98.2)	.71
Yes	1 (1.2)	5 (1.8)	
Postoperative seroma fluid discharge			
No	27 (32.1)	157 (55.1)	**<.01**
Yes	57 (67.9)	128 (44.9)	
Postoperative hair removal			
No	55 (65.5)	124 (43.5)	**<.01**
Yes	29 (34.5)	161 (56.5)	
Postoperative sitz bath use			
No	41 (48.8)	105 (36.8)	.13
Yes	32 (38.1)	139 (48.8)	
Unknown	11 (13.1)	41 (14.4)	

Data are n (%).

Several other factors influencing the risk of recurrence have been identified in published literature. Here, increased age and postoperative seroma fluid discharge were identified as independent risk factors for disease recurrence. In contrast, preoperative antibiotic prophylaxis and post-operative hair removal were effective in reducing recurrence. PSD affects the younger population more than other age groups. The mean age of the patients who had recurrence was 20.8 years. In our multivariate analysis advanced age was a significant predictor for disease recurrence. In another study, the likelihood of recurrence was associated with younger age groups.[13]

Obesity is a known risk factor of PSD yet the effect of obesity on recurrence is controversial in the literature. Some studies showed that BMI is related to higher rates of postoperative complication and recurrence.[14,15] In contrast, a long-term prospective study done by Sievert et al. concluded that BMI has no negative influence on the PSD long-term recurrence rate even after 10 and 20 years.[16] Our results support the findings of Sievert et al that BMI does not correlate with recurrence.

Family history is a non-modifiable risk factor of PSD: 35.5% of our patients had family history, but this finding did not influence disease recurrence. In contrast, a positive family history for PSD was a significant risk factor for recurrence in some previous studies.[8,17]

The chance of recurrence was one-fold higher in patients with a history of prolonged sitting more than 6 hours. Although an increase in risk of recurrence was observed, it was not statistically significant and could have been due to recall bias when patients might inaccurately remember their sitting time. In comparison to a similar Saudi study, these results revealed that prolonged sitting more than 6 hours did significantly affect disease recurrence.[13] The literature has also shown that prolonged hours of sittings affects the recurrence of disease. The groups who are at high risk of recurrence of PSD are those who have prolonged sitting jobs such as secondary school students, soldiers, and drivers.[5,18,19]

To date, various surgical techniques have been proposed for the treatment of PSD but there is no consensus on the gold standard treatment as none of them managed to prevent recurrence completely.[10] Siwei Bi et al conducted a systematic review and network meta-analysis with more than 5000 PSD patients and concluded that the Modified Limberg flap and off-midline

Table 4. Multivariate regression analysis to determine independent factors associated with pilonidal sinus disease recurrence (n=369).

Factor	B	S.E.	Wald	*P* value	Adjusted OR	95% Confidence Interval
Age (years)	0.069	0.024	8.364	0.004	1.072	1.023 – 1.123
BMI (kg/m^2)	0.018	0.020	0.775	0.379	1.018	0.978 – 1.060
Antibiotic prophylaxis						
No						
Yes	-0.851	0.301	7.990	Ref **0.005**	0.427	0.237 – 0.770
Postoperative fluid discharge						
No						
Yes	0.929	0.277	11.264	Ref **0.001**	2.531	1.472 – 4.353
Postoperative hair removal						
No						
Yes	-0.841	0.270	9.669	Ref **0.002**	0.431	0.254 – 0.733

Overall model test: *P*<.001, Deviance: 352.069, Nagelkerke R^2: .170

closure were associated with the lowest recurrence and complications rates, while primary midline closure, which was used in our study, was associated with the highest rate of recurrence.[20] Another meta-analysis focusing on the impact of geographic distribution on recurrence rate found that primary asymmetric closure and various flap techniques were associated with a low recurrence rate regardless of the geographical region. In addition, minimally invasive procedures such as video-assisted endoscopy, phenol injection and marsupialization are also emerging as alternative procedures.[12]

The surgical techniques used in this study were divided into two groups based on closure types (primary or secondary). Our analysis suggests that there was no significant difference in recurrence rate found between the types of closure. This finding is in sharp contrast with a meta-analysis of 26 RCTs including 2530 patients which concluded that leaving the wound to heal by secondary intention was associated with a lower recurrence rate compared to primary closure.[21] However, with small sample size, caution must be applied, as our finding might be prone to type II error. When investigating time to recurrence, primary closure was associated with early recurrence compared to secondary healing. The early onset of recurrence after primary closure could lead to increased reporting of recurrent cases after primary closure compared to secondary healing.

Because of the lack of published studies, the effect of preoperative prophylactic antibiotics on disease recurrence is still controversial. Two randomized control trials demonstrated that prophylactic antibiotics did not appear to be significant in reducing recurrence rates.[22,23] In another randomized control trial, there was a trend towards a lower recurrence rate in the group receiving prophylaxis (6/73 vs. 14/72, *P*=.09) at a late follow- up (>6 years).[24] In our study, we found that preoperative intravenous prophylactic antibiotics were protective against recurrence. In order to further elucidate the effect of antibiotic prophylaxis on recurrence, large, double-blind, placebo-controlled randomized trials are needed.

Analysis of postoperative wound complications showed that postoperative seroma fluid discharge was associated with an increased risk of recurrence. Multiple studies reported seroma fluid as a postoperative complication: 15.1%, 8.6%, and 5.3%.[25-27] However, none of these studies linked seroma to disease recurrence. On the other hand, wound infection and dehiscence had no effect on disease recurrence in our study but they were risk factors for recurrence in another study.[28] Nevertheless, the small number of patients who experienced wound infection and dehiscence in our study warrants caution in interpreting the results because they are prone to significant type II error.

It has been suggested that a high density of body hair at or around the natal cleft may contribute to PSD recurrence.29,30 Our result agrees with these previous results. Here, patients who failed to keep the area hairless after surgery had a higher rate of recurrence. However, we did not investigate the effect of different hair removal methods on recurrence. Several studies have suggested that laser hair removal helps to decrease risk of disease recurrence and should be considered as an adjunct therapy to the surgical treatment to minimize the disease recurrence.[31,32] Pronk et al conducted a systematic review of 14 studies to evaluate the effect of hair removal methods on recurrence rate and showed a recurrence rate of 9.3% in patients treated with laser hair removal compared to no hair removal (19.7%) and razor and cream depilation (23.4%).[30]

Our retrospective study design relied on physician recordings which can limit the availability of some data, but this did not affect statistical analysis significantly. The follow-up time was short, which may underestimate disease recurrence. In conclusion, the ability to inform our patients about the chance of recurrence and its risk factors is still challenging. In this study, the PSD recurrence rate was 22.8%. Advanced age and post-operative seroma fluid were significant independent risk factors for PSD recurrence. Preoperative antibiotic prophylaxis and hair removal after surgery decreased recurrence. Primary closure was associated with early onset of recurrence compared to secondary closure.

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