Int Surg 2021;**105:**30–35
DOI: 10.9738/INTSURG-D-15-00219.1

Pili Multigemini Is a Possible Risk Factor for Pilonidal Sinus Disease

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The aim of this study is to analyze both previously proposed and new risk factors for the development of pilonidal sinus. This is a prospective case-control study consisting of 145 patients with pilonidal sinus disease (n = 45) and a control group (n = 100). All patients were admitted to the department of general surgery between January 2013 and May 2015. The patients' age, family history, medical history, sitting time in a day, sitting posture, body mass index (BMI), Garn hairiness score, and hair type were evaluated. There were significant differences between the groups in the following characteristics: age ($P = 0.01$); positive family history ($P = 0.01$); medical history ($P = 0.01$); sitting time in a day ($P = 0.01$); sitting posture ($P = 0.01$); BMI ($P = 0.01$); Garn score ($P = 0.01$); and hair type. Multivariate logistic regression analysis indicated that positive family history ($P = 0.03$); Garn score ($P = 0.05$); medical history ($P = 0.01$); and sitting posture ($P = 0.02$) were independent risk factors for the development of pilonidal sinus disease.

Key words: Pilonidal sinus – Hair follicle – Pili multigemini

Pilonidal sinus is a common health problem that usually has an acquired etiology and is mainly encountered in young people.[1] The precise etiology of the disease is still unclear. A widely accepted theory is the rupture of a follicle in the natal cleft leading to a sinus containing hairs.[1] This is consistent with the clinical observation that pilonidal patients are often hairy persons and that pilonidal sinus disease (PSD) rarely occurs in populations with less body hair.[1] PSD is basically a disease of the skin, subcutaneous area, and pilosebaceous unit.

High body mass index (BMI) has been identified to be a risk factor for the development of PSD.[2] The sex hormones produced in puberty are known to affect the pilosebaceous gland, which coincides with the earliest onset of pilonidal disease.[1–4]

The hair growth patterns of men were published by Garn[5] in 1951, and the visual scoring system originating from Garn's study evaluates those patterns. In this study, 2600 body-build photos of men are evaluated, and 239 white men are visually examined. The study evaluates the amount of hair

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in 10 different body regions using a 5-point scale (0–4).

To the best of our knowledge, hair type has never been analyzed as a possible risk factor for pilonidal disease. Pili multigemini (PM) involves the structure of hair and was described in the mid-1800s as involving more than one hair shaft originating from the same follicle.[6] Other types of hair anomalies have recently been reported.[7]

Other factors associated with PSD are increased sweating, local irritation, or local trauma.[8] The main aim of this study is to assess the effect of hairiness, hair type, sitting time, and sitting posture along with previously described risk factors for developing PSD.

Patients and Methods

A total of 45 male patients with pilonidal disease were admitted to the general surgery outpatient clinic between January 2013 and May 2015. The control group was composed of 100 age- and sex-matched patients admitted to the outpatient clinic without known PSD, malignancy, or morbid obesity. Physical examination of the presacral region was performed in the control group to identify any possible PSD presentation. Patients discovered to have asymptomatic PSD at the initial examination were excluded from the control group. The patient age in both groups was between 18 and 45 years. Data were collected prospectively. The data collected from each patient included age; family history; medical history (including acne, folliculitis, hidradenitis suppurativa, and recurrent PSD); time spent sitting in a day; types of sitting posture; BMI; Garn score; hair type; and complaints. Sitting time was recorded as >6 hours or <6 hours and sitting posture types were defined as forward sitting (forward leaning sitting); middle sitting; and backward sitting (backward leaning sitting).

Patients were categorized as obese (BMI $\geq$30, n = 7); overweight (BMI range: 25–29.9, n = 35); or normal weight (BMI range: 18.5–24.9, n = 17).

Patients were categorized according to their hairiness scores using the Garn scale, by which hair growth is analyzed in 10 androgen-sensitive areas. The grade for each area ranges from 0 (no terminal hair) to 4 (frankly virile). The body areas analyzed to grade hairiness scores included the beard and moustache, hypogastric, thoracic, lower arm and leg, upper arm and leg, gluteal, lumbosacral, lower back and upper back, and mid-phalangeal regions. Based on this study, total scores less than 5 represent "hairless" and scores higher than 18 represent "hairy" men.[5]

Hair types and disorders were defined as unigeminate, multigeminate, circled, rolled, and ingrown hair. All of the body parts that were visually scored were examined with a magnifying glass (×3) to identify distinct hair types.[6,7]

Comparison between the 2 groups was made using the Student's *t*-test for parametric data and the Mann-Whitney *U*-test for nonparametric data. Comparison of categorical variables was performed using the χ^2 test. Multiple logistic regression analysis was used to define the risk factors of the outcome variable (pilonidal sinus). Values of $P < 0.05$ were considered statistically significant.

Results

Age

The mean age was 23.9 ± 4.5 years (range: 18–43) in the PSD group and 27.1 ± 6.2 years (range: 18–41) in the control group ($P = 0.001$).

Family history

A total of 15 (33%) patients had a positive family history of PSD in first-degree family members, and 30 (66%) patients in the PSD group had no PSD family history. There were only 10 (10%) patients with a family history of PSD in the control group. Positive family history of PSD in first-degree family members was significantly higher in the PSD group than in the control group ($P = 0.001$).

Medical history

History of acne and folliculitis was positive in 36 (80%) patients in the PSD group and 14 patients in the control group. Positive medical history was found to be significantly higher in the PSD group than in the control group ($P < 0.001$). There was a 24% rate of disease recurrence in the PSD group.

Time spent sitting in a day

The mean time spent sitting in a day was 6.2 ± 1.5 (range: 3–10) hours for the PSD group and 3.8 ± 1.7 hours (range: 2–10) for the control group ($P < 0.001$). In total, 71% of the patients in the PSD group and 14% of the patients in the control group spent more than 6 hours sitting in a day.

Table 1 Comparison of the numeric risk factors between the groups

Parameter	PSD, median (min–max)	Control, median (min–max)	*P*
Age, y	23.9 (18–43)	27.1 (18–43)	0.001
Sitting time per day, h	6.2 (3–10)	3.8 (2–10)	0.001
BMI, kg/cm^2	25.4 (19.1–33.8)	22.6 (18.1–30.8)	<0.001
Garn score	27.2 (18–40)	22 (14–31)	<0.001

Table 2 Comparison of the categorical risk factors between the groups

Parameter	PSD n (%)	Control n (%)	Total n (%)	*P*
Family history				
Positive	15 (33.3)	10 (10)	20 (17.2)	0.001
Negative	30 (66.7)	90 (90)	120 (82.8)	
Medical history				
Positive	36 (80)	14 (14)	50 (34.5)	<0.001
Negative	9 (20)	86 (86)	95 (65.5)	
Sitting posture				
Forwards	0	27 (27)	68 (46.9)	<0.001
Middle	6 (13.3)	62 (62)	50 (34.5)	
Backwards	39 (86.7)	11 (11)	92 (63.4)	
Pili multigemini				
Positive	45 (100)	47 (47)	92 (63.4)	<0.001
Negative	0	53 (53)	53 (36.6)	

Sitting posture

In the PSD group, 39 (86%) patients had a backward sitting posture and 9 (13%) patients had a middle sitting posture. In the control group, on the other hand, 27 (27%) patients sat with a forward posture; 62 (62%) patients sat with a middle posture; and 11 (11%) patients sat with a backward posture. Backward sitting posture was significantly higher in the PSD group than in the control group ($P = 0.008$).

BMI

Based on BMI scores, in the PSD group, 16 (35%) patients were normal weight; 22 (48%) patients were overweight; and 7 (15%) patients were obese. In the control group, however, 67 (67%) patients were normal weight; 30 (30%) patients were overweight; and 3 (3%) patients were obese. The mean BMI scores were 25.4 ± 3.9 (range: 19.1–33.8) and 22.6 ± 3.2 (range: 18.1–30.8) in the PSD and control groups, respectively ($P < 0.001$).

Garn score

The Garn hairiness score was significantly higher in the PSD group [27.2 ± 4.7 (range: 18–40)] than in the control group [22 ± 4.2 (range: 14–31); $P < 0.001$]. All patients in the PSD group were above the Garn hairiness cutoff level, and only 25 (25%) patients were under this cutoff level in the control group.

Hair type

With respect to hair type, all patients in the PSD group had multigeminate hair. Among them, 39 (86%) patients had bigeminate hair; 6 (13%) patients had trigeminate hair; and 13 (28%) had rolled and multigeminate hair. In the control group, on the other hand, 53 (53%) patients had unigeminate hair and 47 (47%) patients had multigeminate hair. The multigeminate hair type was significantly higher in the PSD group than in the control group ($P < 0.001$; Tables 1 and 2).

Complaints

The complaints at presentation for this series of 45 patients were as follows: 20 (44%) patients had abscess formation; 19 (42%) patients had purulent discharge; 4 (8%) patients had sacrococcygeal pain; and 2 (4%) patients had bloody discharge. There was recurrent PSD in 11 (24%) patients, and the remaining 33 (73%) patients were diagnosed with PSD for the first time. Multivariate logistic regression analysis indicated that positive family history ($P = 0.03$); Garn score ($P = 0.05$); medical history ($P = 0.01$); and sitting posture ($P = 0.02$) were independent risk factors for the development of PSD.

Discussion

PSD, which was first described by Hodges in 1980,[9] is a potentially devastating pathology affecting patients worldwide. PSD infections and chronic pilonidal sinuses typically occur in the midline of the sacrococcygeal skin of young people.[1] Although the exact pathogenesis of PSD remains elusive and controversial, hair seems to play a central role in the process of infection and in the perpetuation of granulation tissue in sinuses. Treatment of PSD is based on clinical presentation rather than etiologic factors. Lack of information on the exact etiology of PSD causes healing problems and recurrence. In those cases, the treatment performed may not have been fully successful or was insufficient, representing a waste of effort and increased cost. A more effective treatment for the disease will be possible if the real risk factors for the development of PSD can be identified.

PSD is infrequent in black and East Asian people[1,10]; the patients in both study groups were white males. The prevalence of positive family

history for PSD in first-degree relatives has been reported to be 12% to 38%.[14–16] Positive family history was similar between the PSD and control groups of a study reported by Harlak *et al*.[11] In the present study, positive family history for PSD was 33% in the PSD group, which was significantly different from that of the control group. The higher incidence of family history in the study may be due to the incidence of recurrent disease in the PSD group.[12]

The prevalence of family history of PSD was significantly different between groups. These findings suggest that congenital differences may predispose a patient to developing PSD.

All of the patients in both groups were male and of a similar age range. Sex hormones first produced at puberty are known to affect the follicular unit, which coincides with the earliest onset of PSD.[13,14]

The structure of the follicular unit consists of terminal and vellus follicles inserting into the arrector pili muscle and associated sebaceous lobules. In both sexes, sebaceous gland growth and secretory activity increase concomitant with high blood androgen levels. Increased sebum excretion is the main reason for acne and folliculitis.[14] Sebum secretion is less common in women than in men, and secretion is greatly decreased after age 50.[14] PSD is less likely to be observed in women and after age 50. When looking at the medical history of patients with PSD, we see that skin disorders such as acne and folliculitis are concomitantly reported to a high degree. As a matter of fact, it is suggested that persistent folliculitis is one of the reasons for PSD recurrence.[8,15] We did not identify any cases of hidradenitis in either group. In the PSD group, 11 (24%) patients had recurrent disease. We therefore found that all recurrent patients had a medical history of folliculitis. The causative factor for recurrent PSD is probably recurrent infection that is unresponsive to antibiotics.

One of the predisposing factors for developing PSD is time spent sitting in a day, which is why the condition was first described as "Jeep disease."[16] The effect of sitting time in a day on the development of PSD is generally accepted.[10,11] Daily sitting time was significantly higher in the PSD group than in the control group.

Sitting posture is an important occupational factor. The weight of the body is transferred to the ischial tuberosities of the pelvis, sacrococcygeal region, and surrounding soft tissues. In this study, the backward position was predominantly found in the PSD group. In a backward sitting position, body weight is practically slumped down to the sacrococcygeal region and floor support is less than 25%.[17] In such a position, the sacrococcygeal skin and fat tissue are under compression and pressure. The level of this pressure varies according to body weight. Overweight persons are naturally exposed to increased pressure compared with normal weight persons. Furthermore, the backward sitting position causes increased sweating at the sacrococcygeal region.

Obesity has been suspected to be a risk factor for the development of PSD.[2,8] However, the effect of BMI on the formation of PSD is still controversial.[2] It has been reported that high BMI scores increase PSD recurrence.[18] In addition, dietary habits affect sebaceous gland function and sebum composition.[19,20] Increased dietary fat or daily carbohydrate consumption causes increased sebum discharge.[20] Dietary factors are related to dermal disorders.

Obesity causes hormonal, metabolic, and multisystemic disorders, including skin disorders. The changes include problems with skin barrier function, sebaceous glands, sweat glands, the lymphatic system, wound healing, and subcutaneous fat.[21] Obesity increases the incidence of cutaneous infections, including folliculitis.[22] Acne formation is related to overactivated mammalian target of rapamycin complex 1.[23] Obesity is associated with impaired angiogenesis and chronic low-grade inflammation. Increased intracellular glucocorticoids suppress angiogenesis, and hypoxia-inducible factor 1 alpha levels increase in fat tissue. As a result, local chronic inflammation and fibrosis develop. Obesity decreases adiponectin levels in fat tissue. Adenosine monophosphate–activated protein kinase signaling and the extracellular signal–regulated kinases signaling pathway do not work properly, which results in impaired perfusion of tissue and the re-epithelialization phase of the wound healing process.[24] However, wound healing results are reported to be similar in obese and normal weight patients.[4]

Ethnicity and racial variations affect the frequency of hair follicles and hair growth. East Asian males were reported to have fewer follicles per skin area than Euro-American (white) males of a similar age.[25] It is possible that ethnic and racial variations in body hair growth have an impact on the scores and prevalence of hairiness in males. PSD frequently develops in hairy populations or persons.[8,11] However, no objective data have been analyzed using a visual scoring system in the literature. To the best of our knowledge, the Garn visual scoring system was used for the first time in this study. The cutoff values

reported by Garn are different than those used in the present study. The male hairiness rate in Turkish society is reportedly higher than the values reported by Garn. In the present study, the Garn scores of both groups were higher than the scores reported in the original study, and all patients in PSD group had scores above the cutoff values.

Hair characteristics vary by body part. The hairs in pubic and axillary areas are in vellus form in the first years of life. During puberty, the hairs in these areas become terminal hairs due to growth hormones and androgens. The real cause of hair type disorders is not known; they may be the result of androgen levels or genetic factors. Pili multigemini is a distinct type of hair growth in which two or more hair strands grow from one follicular ostium. The diagnosis of pili multigemini is primarily made visually, and the condition is reported to be rare (2%).[26] However, in a study composed of 30 male and female patients, pili multigemini was present in all subjects.[27] Pili multigemini is associated with folliculitis, and tufted hairs in particular are reported with recurrent inflammatory papules, leaving atrophic/hypertrophic scars.[28] The results of our study were not consistent with those reported in the literature as all patients in the PSD group had hairs with PM characteristics; additionally, the same patients had rolled hairs. The cause of PM could be genetic or silent embryonic epithelial germ reactivation in adolescents.[29]

Embryogenetic development of hair placodes is affected by the β-catenin (Wnt signaling) pathway.[30] After birth, hair shaft differentiation is guided by stem cells placed at the shaft.[31] Chamberlain and Vawter reported that natal cleft biopsies showed 8% of children to have tracts or remnants of tracts associated with PSD.[32] This result is 10-fold higher than the reported prevalence of asymptomatic PSD found in the general population.[12,33] Human tissues encounter many mechanical forces, one of which is compression. These forces guide tissue growth and differentiate mesenchymal stem cells to form mature tissues.[34] There is enough evidence reported in the literature to suggest that outer mechanical forces determine the fate of stem cells.[35] We may suggest that repetitive mechanical compression to the sacrococcygeal region may affect the growth and differentiation of epidermal stem cells, which are placed in the congenital natal cleft tract.

The complaints of the PSD patients at presentation were mostly abscess formation and purulent discharge. Abscess formation was the most frequent complaint, which is consistent with the literature.[8]

Conclusion

These results indicate that positive family history, positive medical history, long sitting periods, and hairiness are proposed risk factors for the development of PSD. Additionally, this study suggests that pili multigemini and backward sitting posture are new risk factors for PSD. There may be a genetic cofactor similar to PM, as PSD is associated with familial predisposition. Further studies with a larger sample size would be more predictive for determining new PSD risk factors.

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