



Unusual sites and presentations of Pilonidal Sinus Disease

Gupta Pravin J, Gupta Shalini M, Heda Purushottam S.
Fine morning hospital and research center, Laxminagar,
Nagpur,INDIA

rezime Pilonidal sinus usually develops in the sacrococcygeal area or other hair-bearing areas. However, it can present at some other sites with varied presentations and can mimic like other infective pathologies which are more common in that area. It has also been described as an occupational hazard in barbers, especially when presented interdigitally. Occupational pilonidal sinuses tend to occur even in non-hair-bearing areas where there is no presence of individual's own hair. Pilonidal sinus occurs in many areas of the body such as web of fingers, penis shaft, axilla, intermammary area, groin, nose, neck following trauma during shaving, clitoris, sternum, suprapubic area, occiput, prepuce, chin, periungual region, breast, face or navel. Sporadic incidences of implantation of pilonidal sinus have been reported. The disease is mostly observed in hairdressers, but it has also been reported sporadically in other professions, as male sheep shearer, dog groomer, slaughter men or milker of cows. Short customers' hairs that penetrate the supple interdigital skin of the hands produce barber's disease. Though there are only a few reports about these peculiar sinuses in the available literature, they should be suspected in any chronically discharging, non-healing sinus or wound. A clinical suspicion of malignant transformation or affection with tuberculosis in the endemic area should also be kept in mind to avoid any delay in the treatment and the outcome of the disease.

Key words: pilonidal sinus, site, presentation

INTRODUCTION

Pilonidal sinus of the interdigital spaces of the hand is a well-recognized occupational disease of male barbers¹. The higher incidence of the disease in male hairdressers has been attributed to those female hairdressers seems to be more diligent as concerns the cleansing of

the interdigital spaces of the hands and feet²). The interdigital spaces are susceptible to penetration by hair because the epidermis is very thin in this area; it is easily irritated by moisturizing agents and shampoos routinely used by hairdressers, while the tile-like formation of the cuticle can act as a barbed hook. Furthermore, clipped hairs are sharp as a needle, moist, electrostatic, adhesive, and preferably accumulate in the web spaces.

The exact reasons for the lesion are not known. However, several theories are offered. Hair penetration, negative pressure from finger abduction, recurrent infection and chronic infection are considered factors involved in the establishment of an interdigital web space sinus.

The lesion is produced by the penetration of foreign-born short hairs into the interdigital spaces of the hand. The hairs produce an inflammatory reaction and foreign body granuloma. They cause a sinus, and later a cyst³.

Through the sinus, the hairs are entrapped and may occasionally be expressed. Moreover, chronic, purulent drainage may occur. The structure of the lesions varies from epithelial-lined tract, cyst with surrounding foreign body reaction, to fibrotic cicatricial tissue. Most sinuses are asymptomatic and individuals may not even be aware of them. Although the clinical picture is usually benign, it can be complicated by repeated infection, which may require surgery. Abscess formation, cellulitis, lymphangitis and osteomyelitis are possible complications of barber's hair sinus.

A synonym for pilonidal sinus is pilonidal granuloma. The histopathological appearance of the lesion is characteristic of a foreign body granuloma. An epithelial-lined sinus tract leads to an area of fibrosis and granulation tissue surrounding hair shafts.

Although thorough removal of imbedded hair might result in complete cure of the condition in certain cases, conservative measures in symptomatic sinuses have not

actually proved to be very effective. Dorsal metacarpal artery perforator flap is another choice with minimal donor site morbidity and which can provide robust skin coverage to avoid further penetration of hair in to web space.

Careful cleansing and drying of the interdigital spaces, as well as use of protective barrier creams, adhesive band-aid type strips, collodion, or fingerless gloves, which maintain pulp sensitivity could prevent the formation of the disease⁴. Moreover, hairdressers are advised to wear socks and shoes that do not expose the feet in order to prevent the formation of a pilonidal sinus on the feet. However, the main preventative method is the careful removal of any hairs that have penetrated the epidermis at the end of the working day.

UMBILICAL PILONIDAL SINUS (UPS)

Pilonidal sinus disease of the umbilicus is caused by hair penetrating the skin, causing a foreign-body reaction and development of a sinus lined by granulation tissue. Umbilicus could be an ideal area for PS formation since it is a depressed, moist and hairy area. Patients may not be symptomatic initially, but most complain of pain, discharge or bleeding at the umbilicus when symptoms do develop. With good lighting conditions and the help of an assistant to retract the skin of the umbilicus, hairs can be seen deep in the umbilicus and usually protrude from a small sinus. Additional diagnostic procedures are usually not necessary. Deep navel, an important anatomic variation, is common in such patients, which indicates a strong correlation with the disease. Additionally, inadequate personal hygiene is also noticed in the majority of patients. UPS occurs more frequently in young hairy males and hence is more common among students⁵. The fact that young males prefer tight clothes may lead UPS to occur more commonly in this age group. Being hirsute is probably the most important predisposing factor. A strong family history of UPS was also noted. Wearing belt causes the hairs to be collected at the level of umbilicus and sets the ground for a moist environment, with the hairs piercing the skin. It is also thought that taking bath infrequently allows the hairs to be accumulated in umbilicus, inducing the development of UPS.

Simple extraction of hair from the sinus will relieve symptoms in most patients. Occasionally, incision and drainage of an abscess may be necessary. More aggressive surgical therapy should be used only after conservative management has failed. Surgical procedures in which umbilicus is completely removed, may cause cosmetic losses. Furthermore, losing the umbilicus may give rise to psychological misperceptions and make one feel as if he/she has lost connection with the mother, ancestors or even humanity. In order to avoid the psychological effects of such procedures, detection of etiological factors of UPS and determination of methods for preventing them would be more useful.



FIGURE 1
TUBERCULAR PILONIDAL SINUS

PILONIDAL SINUS OF THE PENIS

Pilonidal sinus of the penis is a rare entity, with very few reported cases. They clinically present as a classic case of inflammation with pain, local infection, and redness, but may also show chronic ulceration or a draining sinus or abscess formation. Pilonidal sinus of the penis is a rarely reported entity in uncircumcised men, the most common site being the region around the corona involving the foreskin for pilonidal sinus to occur in the penis⁶. It is hypothesized that the coronal sulcus acts as a cleft where hair may accumulate from surrounding hairy areas of the patient or possible partner. The hairs are then driven into the shaft and prepuce by the mechanical forces and the rolling movement at the junction of glans penis and the uncircumcised prepuce⁷. Simple excision with primary closure or healing by granulation tissue and sinus tract excision is usually the treatment for pilonidal sinus. If pus is available, it should be sent for culture and Gram stain. Long-term penicillin is prescribed after cultures and antibiotic susceptibilities to prevent sepsis⁸. This is followed by circumcision to treat the underlying phimosis. In conclusion, pilonidal sinus should be considered an important differential diagnosis in a case of penile swelling, non-healing ulcer, or phimosis.

INTERMAMMARY PILONIDAL SINUS

Intermammary pilonidal sinus disease is commonly seen in fatty females with increased distribution of hairs. After the onset of puberty, sex hormones affect the pilosebaceous glands, and, subsequently, the hair follicle becomes distended with keratin. As a result, a folliculitis is created, which produces edema and follicle occlusion. The infected follicle extends and ruptures into the subcutaneous tissue, forming a pilonidal abscess. This results in a sinus tract that leads to a deep subcutaneous cavity. The direction of the sinus tract is cephalad in 90% of the cases, which coincides with the directional growth of the hair follicle. The laterally communicating sinus is created as the pilonidal abscess spontaneously drains to the skin surface.

The original sinus tract becomes an epithelized tube. The laterally draining tract becomes a granulating sinus tract opening.

The sinus is caused by the friction of the skin leading to the embedding of the hair beneath the surface. The hair forms small cavities or pits, which are in truth, enlarged hair follicles, which go on to become sinuses. Bacteria and debris enter this sterile area, producing local inflammation and formation of pus-filled abscesses. In chronic condition, the sinus becomes an open cavity, constantly draining small amounts of fluid.

Although intermammary pilonidal disease may manifest as an abscess, pain and purulent discharge are the two most frequently described symptoms. In the early stages, only a cellulitis or folliculitis is present. The abscess is formed when a folliculitis expands into the subcutaneous tissue or when a pre-existing foreign body granuloma becomes infected⁹

The subcutaneous cavity and laterally oriented secondary sinus tract openings are lined with granulation tissue, whereas only the midline natal cleft pit sinus is lined by epithelium. The diagnosis of a pilonidal sinus can be made by identifying the epithelized follicle opening, which can be palpated as an area of deep induration beneath the skin. Treatment for symptomatic sinus involves surgery to incise and drain the abscess. The surgery can be either wide excision and healing by secondary intention, excision and primary closure by sutures, or plastic surgery technique.

AXILLARY PILONIDAL SINUS

Sporadic reports of a pilonidal sinus in the axillary region have also been reported. This usually occurs in a hirsute person with the history and complaint of the intermittent small amount of leakage from axilla. Friction (abduction – adduction), suction, massage, shaving, pounding, minor infection and maceration are assorted mechanisms which play a part in causation of pilonidal sinus¹⁰. On physical examination, a single or occasionally multiple sinuses, which may or may not include any hair, are noted. The sinus may or may not be expressing any discharge. The area around the sinus is edematous. Total excision with primary closure is reportedly an ideal treatment in this situation¹¹.

PILONIDAL SINUS OF THE EYELID

Pilonidal Sinus over eyebrows is very rare. It can present as a skin tag-like protruded lesions over the eyelids. The presenting symptoms are itching and pain over the eyelid. Clinically, it can present as a pustule adjacent to the protrusion. The possible cause of this site with pilonidal disease could be due to regular plucking and shaping of hairs by threading of eyebrows. The protruding hair from the sinus can be captured using forceps and the complete tract can be visualized. The sinus tract can be dissected superficially by radiofrequency or electrocautery in assistance with an ophthalmic surgeon¹².

It can be hypothesized that the routine procedure of plucking of eyebrows may initiate the suction force, which is required for pathogenesis. The wrinkled skin present over eyelids could have contributed to the penetration of hair.

PILONIDAL SINUS OF THE NOSE

Three cases of pilonidal sinus of the nasal pyramid have been reported so far in world literature. Concerning the nose, it is obvious that any negative suction as like in other types of sinuses cannot be created over the nasal pyramid. It was suggested that pilonidal sinus of the nasal dorsum is a congenital lesion. If incompletely excised during childhood, it may recur in adulthood with symptoms of pilonidal disease or, the previous lesion could have been a nasal dermoid, the excision of which led to implantation of epithelium and development of a pilonidal sinus. Pilonidal sinuses developing in such sites, due to implantation of hair in diseased or surgically traumatized skin has been described before too. On the nasal dorsum, a diagnostic dilemma exists between a pilonidal sinus and dermoid cyst. Dermoid sinus cysts appear within eighteen months of birth, while pilonidal sinuses are adult onset in origin¹³. Both have hairs protruding from the sinus opening, but extrusion of cheesy material is seen only in the dermoid cyst. The histopathology gives the final diagnosis in this issue. The dermoid is characterized by the presence of adnexal structures like sweat glands, sebaceous glands and hair follicles, which may still have hairs attached to them, whereas the pilonidal sinus is not associated with adnexal elements and has fragmented hair shafts lying loose within its lumen.

TUBERCULAR AFFLICTION IN THE PILONIDAL SINUS

Tuberculosis is a broad-spectrum disease that may involve pulmonary and extra pulmonary locations. Tuberculosis (TB) is a major public health problem, affecting 8 million persons per year worldwide. The global incidence rate of TB per capita is growing by ~1.1% per year. Contrary to the increasing number of TB cases in developing countries, the number of cases in industrialized countries is stable or decreasing. Nevertheless, a decreasing trend of the total number of TB patients is seen with an increasing proportion of TB cases with extra pulmonary TB. Both the HIV epidemic and changes in population demographics, with rising numbers of immigrants, are being held responsible for this proportional increase of extra pulmonary TB. Extra pulmonary tuberculosis is responsible for 15% of all cases of tuberculosis. In many countries, patients from Asian origin are known to have a higher incidence of extra pulmonary TB. Tuberculous infections have been increasing in incidence during the last decades for a variety of reasons, including increasing numbers of patients with immunity-depressive diseases, drug resistance, aging population, and health care worker exposure. As the rate of patients with extra pulmonary tuberculosis has increased globally in the last few years,

the perianal localization is also increasing in similar proportion. Tuberculosis should be suspected in patients with complex or recurrent perianal septic lesions. The most frequently encountered perianal tuberculous lesions are suppurations and sinuses (Fig.1).

It is difficult to explain the possible cause of tubercular affection in the pilonidal sinuses. Cutaneous tubercular abscess can occur from extension of an embolism to subcutaneous tissue (such as pulmonary foci or direct skin inoculation) or from extension of an underlying lymphadenitis, synovitis, or osteomyelitis¹⁴. TB has also been described following subcutaneous or intramuscular injection. Either the syringe, needle or fluid to be injected has been contaminated or the medical attendant has exhaled tubercle bacilli into the patient's skin, which are then introduced by the injection. It may be due to a direct inoculation from the stool of the patient, which may be containing tubercular bacilli. Another possibility is that the pre-existing sinuses are infected with tubercle bacilli either by way of finger or by the use of toilet paper. As tuberculosis in the pilonidal sinus is rarely diagnosed before operation based on the clinical picture, the histological examination of the tract of the sinus is mandatory for the correct diagnosis. Novel diagnostic modalities such as adenosine deaminase levels and polymerase chain reaction can be useful in doubtful situations.

There should be a strong clinical suspicion of tuberculosis in endemic areas with such presentations as *Mycobacterium* one of the causes of granulomatous diseases of the skin and subcutaneous tissues¹⁵. Patients with such presentations are treated several times in the past by the family physicians considering it as boils or abscess. On occasions, it is squeezed and drained and at other times, it may be treated with antibiotics. The treatment often results in arresting the symptoms for the time being, but would recur after few weeks with similar symptoms and presentations.

Treatment of tubercular pilonidal sinus disease included two parts: conventional surgical treatment of sinuses and specific medical antituberculosis treatment.

Antituberculosis treatment is the mainstay in the management of tubercular sinuses. However, the ideal regimen and duration of treatment have not yet been resolved. Since 1982, the American Thoracic Society and the Centers for Disease Control have recommended a nine-month course of isoniazid and rifampicin for the routine treatment of TB in the United States. However elsewhere, a shorter course of four or six months of chemotherapy can be recommended for the treatment of perianal tuberculosis.

MALIGNANT CHANGES IN THE PILONIDAL SINUS

Common complications of pilonidal sinus include cellulitis, abscess formation, and recurrent sinus development. Less commonly, sacral osteomyelitis and meningitis can occur. Carcinoma arising from pilonidal disease is a rare complication occurring in the setting of long standing inflammation. Local recurrence is common and tends

to occur early. Repeat surgery for recurrent disease may involve extensive resection.

Malignant degeneration occurs in approximately 0.1% of pilonidal sinuses. Males are most often affected, with a mean age at diagnosis of 50 years. The average duration of antecedent pilonidal disease is above 20 years. The mechanism by which malignant degeneration arises in a pilonidal sinus is believed to be the same as for other chronically inflamed wounds, such as scars, skin ulcers and fistulas.

Immunosuppression and human papilloma virus infection may be predisposing factors to malignant degeneration of pilonidal cysts and may accelerate the transformation¹⁶.

Most of these malignancies are squamous cell carcinoma. Rarely Basal cell carcinoma arising in the pilonidal sinus has been reported.

Pilonidal carcinoma has a rather distinctive appearance, with the diagnosis frequently suspected by inspection, based on the presence of a long-standing and persistent pilonidal sinus with drainage, sudden rapid growth, overgrowth above the skin level, friability, ulceration, hemorrhage in the tissues, external bleeding, and most commonly, bleeding in sinus that has been present for many years

Initial biopsy is to be performed to confirm the diagnosis. CT scanning may be useful in indicating the extent of the local disease and detecting any metastatic spread.

Treatment of choice remains en bloc resection, including the presacral fascia. Wide excision with tumor-free margins is performed with inclusion of skin, subcutaneous tissue, muscle, and, if indicated, portions of the sacrum and coccyx. Surgical treatment has reportedly yielded five-year disease-free states in 55% percent of patients. Closure of the ensuing defect may be accomplished with mesh grafts, split-thickness skin grafts, or vascularized flaps, including gluteal rotation flaps and gluteal advancement flaps.

Some authors propose consideration of adjuvant chemotherapy and radiation to decrease the local recurrence rate. When radiotherapy is added to surgery alone, recurrence rates decrease from 44% to 30%. Re-excision of local recurrence resulted in some long-term survivals.

Few successes have been achieved in patients who were treated with open and thick liquid nitrogen spray (cryosurgery) while monitoring the temperature through thermocouples.

CONCLUSIONS

Pilonidal sinus disease is a common problem of sacrococcygeal region. However, it is also observed in other parts of the body. Though there are only a few reports about these peculiar sinuses in the available literature, they should be suspected in any chronically discharging, non-healing sinus or wound. A clinical suspicion of malignant transformation or affection with tuberculosis in the endemic area should also be kept in mind to avoid any delay in the treatment and the outcome of the disease.

SUMMARY

NEUOBIÈAJENE LOKALIZACIJE I POJAVE
PILONIDALNOG SINUSA

Pilonidalni sinus (PS) se obièno javlja u sakrokokcigealnoj regiji ili drugim mestima obrasli dlakom. Međutim, on se može javiti i na nekim drugim mestima u različitim formama koje mogu da lièe na neke druge infektivne patologije koje su mnogo èešæ na tom mestu.

Ova bolest je takođe opisivana kao profesionalni rizik berbera, posebno kada se javi između prstiju na ruci. PS se može pojaviti èak i na delovima tela koji nisu obrasli dlakom, kao i na mnogim drugim mestima, kao što su: koren prstiju, penis, pazušna jama, između dojki, prepone, nos, na vratu posle povreda za vreme brijanja, klitoris, grudnoj kosti, suprapubiènoj regiji, potiljku, prepucijumu, bradi, oko nokata, na dojicama, licu ili pupku. Saopštavana je i sporadièna pojava implantacionog PS. Bolest se najèešæ sreæ kod frizera, ali ponekad i kod drugih zanimanja, kao što su osobe koje strižu ovce, odgajivaèi pasa, mesari ili muzaèi krava. Kratka kosa mušterija koja probija gipku kožu između prstiju na rukama je uzrok berberske bolesti.

Ključne reèi: pilonidalni sinus, lokalizacija, prezentacija

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