Pilonidal Sinus of the Anal Canal: A Rare Entity - Case Report

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Abstract
Pilonidal sinus is the term first used by Hodges in 1880 to describe a granulomatous lesion containing hairs. It has been previously described as hair extracted from an ulcer and referred to as Jeep’s disease, in world war days. Pilonidal sinus is the condition in which a sinus or fistula is situated at a short distance from the anus and generally contains hairs. It is commonly observed in the sacrococcygeal region and a few other sites, such as the axilla, umbilicus, face, etc. Its presence elsewhere is uncommon. Predisposing factors are traumatized, hairy skin, which leads entry of the hair inside the wound, thus forming a sinus. A 31-year-old patient presented with discharging endoanal sinus tract, which, on exploration, turned out to be an endoanal pilonidal sinus containing hair tufts, a rare case. The patient complained of recurrent pus discharge and anal pain for 45 days, reporting history of travelling around 70 km daily in a two-wheeler vehicle.

On first physical evaluation, a small endoanal bulge was found. It was located at 1 o’clock in lithotomy position (anterior), with purulent discharge and anterior anal fissure. For confirmation, an endoanal ultrasonography (USG) was performed, which showed a sinus tract containing internal echoes and gas bubbles with a small amount of pus. The sinus was explored, and a wide excision was made and left open for secondary healing.

Keywords
► pilonidal sinus
► endoanal pilonidal sinus
► Jeep disease
► driver’s bottom

Pilonidal sinus of the anal canal is a rare entity, and it can be found in hairy patients who are used to going on long, daily rides on two wheeled vehicles.

Introduction
Pilonidal sinus is a disease in which a sinus or fistula develops containing hairs. The term pilonidal sinus was coined by Hodges in 1880.¹ ² It was first described by Anderson in 1847 as “hair extracted from an ulcer”.¹ ³ It is an acquired condition, due to the penetration of hairs into the skin. Clinically, it may be present as a discharging sinus with a small pit on the surface of the skin.

Pilonidal sinus is common in young, hairy individuals. Men are affected more frequently than women. Pilonidal sinus is commonly found in the sacrococcygeal region. Other
sites are the interdigital cleft, axilla, umbilicus, and face. During World War II, pilonidal sinus, was often called Jeep disease because it was more common in soldiers who travelled in a vehicle for a long period.

Pilonidal sinus in the anus is very rare. Only 11 cases have been reported in the medical literature to date. Here, we present a case of a recurrent discharging anal sinus, which was clinically diagnosed as endoanal pilonidal sinus after surgical management.

**Case Report**

In April 2023, a 31-year-old male reported to Chaitanya hospital, Dhule, with a recurrent pus discharge from the anus for 1 ½ month, with itching and pain. He reported history of travelling about 70 km daily on a two-wheeler. On clinical examination, a small bulge in the 12-to-1 o'clock area of the anal verge was observed, with purulent discharge from a fissure at 12 o'clock.

The transanal USG revealed a sinus tract measuring 40 mm in length with internal echoes and gas bubbles, appearing intersphincteric and communicating with the anal canal at 12-to-1 o'clock.

Provisionally, it was diagnosed sinus-in-ano with no external opening.

The patient was subjected to surgery after exploration surgery revealed an endoanal sinus containing a tuft of hairs in its interior and a small amount of pus. Removal of all the hairs and pus was done, followed by complete excision of the sinus tract. The wound was left open to heal by secondary intention. In the postoperative period, the patient had no pain or pus discharge. The patient was followed up for 3 weeks, until the wound healed completely.

**Literature Review**

For many years, pilonidal sinus was thought to be a lesion confined to the postanal region. In 1946, however, Patey and
Scarff reported a case of pilonidal sinus of the finger web in a barber. Further examples of the condition in the hand have been described by Ewing (1947), Patey and Scarff (1948), King (1949), Currie et al. (1953)—who gave a good review of the literature and pathology of this variety of the condition—and by Oldfield (1956). Pilonidal sinus has also been observed in the axilla (Aird 1952), the perineum (Smith 1948), on a midthigh amputation stump (Shoesmith 1953; Gillis 1954), in the umbilicus (Patey and Williams 1956; Clery and Clery 1963, Thorlakson 1966; Colapints 1977), and in the suprapubic region (MacLeod 1951; Patey and Curry roba: Crosby 1962). We do not intend to describe the pathological or clinical features of these forms of the condition.

Different pilonidal sinus presentations are described in the literature. Regarding those located in the endoanal region, 10 cases have been reported in men and 1 in a woman. Regarding age, these were all young patients, 23, 27, 29, 30, 42, 46, 55, 58, 42, and 39 years for the men, and 42 years for the woman.

Six patients presented recurrent purulent discharge and were submitted to the most diverse surgical procedures without clinical improvement. Two of them had no symptoms and were diagnosed during a surgical procedure to correct hemorrhoids. Three of them were symptomatic for the first time. None of the evaluated patients had found hair in the endoanal region by themselves. As in the presented case, there was only 1 sinus in 10 of the reported cases, and 1 of the patients presented 2 endoanal sinuses.

Regarding the surgical procedure, in six cases the cyst trajectory was opened and left to heal by second intention. Just like the case presented here, and on the other five cases, the lesion was completely removed.

In three of the reported cases, the patients were initially surgically treated to correct an anal fistula with seton, which evolved with symptoms recurrence and developed into an endoanal pilonidal sinus.

One of the previous case reports had a 2-year follow-up, and none of the 5 patients presented disease recurrence. Another author presented a 1-year follow-up with recurrence. All other case reports presented no information about the follow-up period.

Discussion

Different pilonidal sinus presentations are described in the medical literature in different sites. Regarding those located in the endoanal region, 11 cases have been reported in the literature to date. The 1st case of endoanal pilonidal sinus was reported in 1971. Pilonidal sinus typically occurs in the 2nd or 3rd decade of life, the average age of the patients is 25 years, and 73.7% are male. As per the cases of endoanal pilonidal sinuses currently reported in the literature, 91% of the cases are in male patients and 9% are in females (10 cases were in males and 1 in a female).

When it comes to the age, those were all young patients 23, 27, 29, 30, 42, 46, 55, 58, 42, 39 years for the men, and 42 years for the woman.

The origin of pilonidal sinus is not well understood. There are two theories for its pathogenesis, congenital and acquired. However, most of the opinions favor the acquired theory.

In general, at least three conditions need to be fulfilled for pilonidal sinus to occur; the first, is ingrown hair in skin; the second is some sort of wrinkled skin, like the natal cleft or the scar; and the third condition is a mixture of hormonal and hygienic problem.

It is an acquired condition, more common in people who have curly or coarse hair because the hairs are curled around and grow back into the skin due to mechanical trauma from bumps, friction, and infection, such as folliculitis. There are several ways to explain the formation of this sinus in the anal canal. Penetration of hair fragment in the subcutaneous tissue through an open anal fissure; the hair may penetrate through the healing wound or newly formed scar after any surgical procedure. Trauma due to riding in bumpy vehicles for long periods is apparently an important factor.

The presenting clinical features of endoanal pilonidal sinus are pus discharge, itching, and pain.

To our knowledge, there are 11 cases reported in the literature, 9 of them presented with recurrent purulent discharge. Two cases were symptomless and were accidentally found during surgical intervention to manage hemorrhoids.

In the case presently discussed, the patient had an anterior anal fissure and reported history of travelling 70 km daily on a two-wheeler.

In World War II days, pilonidal sinus occurred frequently in soldiers travelling in open jeeps for long distances in reclined sitting position, which exposed the natal cleft area and made it susceptible to unhygienic condition and skin breach. But in this case, the patient was travelling in a two-wheeler, in erect posture, for long distances daily, making the anal region and perianal skin susceptible to trauma/skin breach, along with unhygienic condition due to sweating and hairy skin. In this case, there was also a preexisting anterior anal fissure through which coarse hairs might have entered, leading to the appearance of sinus tract, as shown in the transanal ultrasonography.

Conclusion

1) Endoanal pilonidal sinus is a rare entity, with only 11 cases having been previously reported in the medical literature worldwide; this is the 12th case.

2) Hairy, unhygienic skin due to excess sweating around the natal cleft, along with long-distance travel in slightly reclined position, that is, in a four-wheeler vehicle, are the common predisposing factors to this condition in the sacrococcygeal area.

3) Hairy, unhygienic skin due to excess sweating around the anal area, along with long-distance travel in the upright position, that is, on a two-wheeler vehicle, and preexisting anal fissure might be common
predisposing factors in the endoanal pilonidal sinus case described in the present article.

Conflict of Interests
The authors have no conflict of interests to declare.

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