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Etiological Theories, Risk Factors and Recurrence Factors of Pilonidal Sinus

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Abstract: ***Pilonidal sinus is a chronic inflammatory condition that typically occurs in the sacrococcygeal region. Its etiology and pathogenesis involve both congenital and acquired factors. The congenital theories include viewpoints such as sacrococcygeal sacral canal cystic remnants, developmental deformity of the central suture, and glandular degeneration remnants. The acquired theory is mainly based on the penetration theory, supplemented and verified by Karyadaki's formula PD = H × F × V² (where H represents loose hair, F stands for inward attraction, and V denotes damaged skin).***

Keywords: Pilonidal sinus, Etiology, Risk factors, Recurrence factors, Treatment methods.

1. Introduction

The main risk factors for pilonidal sinus include being male, young age, obesity (BMI ≥ 25 kg/m²), poor personal hygiene, thick or hard body hair, proneness to sweating, sedentary habits, a deep gluteal cleft, and a family history. Currently, there are various treatment methods, but the recurrence rate remains as high as 25%-30%. Studies have shown that there is no statistically significant correlation between gender and recurrence. In terms of age, adolescent patients have a higher recurrence rate. Obesity is a contributing factor to the onset, but its impact on recurrence is controversial. Postoperative hair removal in the gluteal cleft (especially laser hair removal) can reduce the recurrence risk. Smoking significantly increases the postoperative recurrence rate and the incidence of wound complications.

Regarding treatment methods, non-surgical treatments such as antibiotic therapy have no significant effect on recurrence, while therapies like phenol injection, fibrin glue, and platelet-rich plasma have their own characteristics. Surgical treatments include incision and drainage, primary suture after lesion resection, secondary healing, and various flap surgeries, etc. The recurrence rates of different surgical methods vary, and there is currently no clear optimal surgical approach. Clinically, the choice should be made based on the patient's condition, wishes, and the surgeon's proficiency.

Pilonidal sinus disease is a chronic inflammatory condition that typically occurs in the sacrococcygeal region, causing discomfort and complications for patients. Research has explored the potential relationship between factors such as obesity, hirsutism, and sex steroids and the development of pilonidal sinus disease. In addition to the typical presentation in the intergluteal cleft, cases of pilonidal sinuses in atypical locations (such as other areas) have also been reported, which can cause symptoms like pain.

Pilonidal sinus in the sacrococcygeal region is a relatively rare benign disease in the clinical anorectal field in China. The mainstream causes and pathogenesis involve congenital and acquired factors. For the congenital theory, there are viewpoints such as sacrococcygeal sacral canal cystic remnants, central fissure developmental deformity, and glandular degeneration remnants. As for the acquired theory, the main one is the penetration theory.

2. Congenital Theory

2.1 Theory of Cystic Remnants in the Sacrococcygeal Sacral Canal

In 1887, F. Tourneax and G. J. Hermann [1] proposed that the coccygeal epithelial channels develop from the remnants of the primitive spinal cord. During embryogenesis, the cranial end (which forms the brain) and the tail end (attached to the skin of the gluteal sulci) of the spinal cord become fixed. In the third month of pregnancy, uneven growth occurs between the spine and the spinal cord, and as a result, the spinal cord eventually thins. During further development, the thinned regions of the spinal cord (terminal filaments) partially indent and partially remain in the shape of grooves lined with cylindrical epithelium. This groove usually disappears by the end of the sixth month of pregnancy. If it fails to close, it may lead to the occurrence of pilonidal sinuses.

2.2 Theory of Developmental Deformity of the Central Suture

In 1878, Fere [2] put forward the hypothesis of central dysplasia in the sacrococcygeal region. Abnormal lesions located deep in the skin of the sacrococcygeal region lead to the formation of inclusion bodies in the soft tissue, resulting in sinus formation and secondary infection, which in turn causes pain, discharge, and cellulitis.

2.3 Theory of Glandular Degeneration Remnants

H. B. Stone (1931) made observations on the urethral gland, a secondary sexual organ in birds. He described the pilonidal sinus as analogous to the urethral gland. These glands are secondary sexual characteristics in birds, and similar structures exist in the ectoderm of the outer ear and breast in humans. This supplements the congenital theory and was supported by Kallet H. I. in 1936. Kallet pointed out that pilonidal sinus disease mostly occurs during adolescence, that is, during the development of secondary sex glands under the influence of pituitary hormones [3].

2.4 Acquired Theory

The main acquired theory is the penetration theory, which is currently widely recognized by scholars. This hypothesis holds that the main cause of pilonidal sinus is that during puberty, with the appearance of secondary sexual characteristics, body hair becomes denser and harder. At the same time, during human activities, the gluteal regions constantly rub against each other, causing skin damage [4]. As a result, the hard hair in the gluteal region is more likely to penetrate the surrounding skin and form tiny channels. In the early stage, the roots of the hair in these channels remain in the original skin, leading to secondary dermoid metaplasia. Later, the metaplastic tissue in the channels generates suction, drawing in hair that has separated from the roots, thereby forming a sinus tract.

Hair entering subcutaneous fat is regarded as a foreign body. When combined with bacterial infection, it may cause chronic cysts or acute abscesses. Therefore, the onset of pilonidal sinus can be sequentially divided into two stages: first, the formation of a penetrating sinus tract, followed by the formation of an inhaled sinus tract [5].

In addition, Karyadaki proposed a pathogenesis theory of pilonidal disease (PD) based on three variables. He identified three major elements leading to the onset of pilonidal sinus: loose hair (H), inward attraction (F), and damaged skin (V). The formula is [6]:

$$PD = H \times F \times V^2$$

This theory has been widely accepted by scholars and has supplemented and verified the acquired theory.

3. Risk Factors for Pilonidal Sinus

The main risk factors for the onset of pilonidal sinus include being male, young age, obesity (BMI $\geq$ 25 kg/m^2), poor personal hygiene, thick or hard body hair, proneness to sweating, sedentary habits, a deep gluteal cleft, and a family history.

4. Recurrence Factors

Currently, there are various surgical methods for treating pilonidal sinus, but the recurrence rate remains as high as 25-30%.

4.1 Relationship between Gender and Recurrence of Pilonidal Sinus

The incidence of pilonidal sinus varies significantly between men and women, and the age of onset in women is generally earlier than in men. The average age of onset for men and women is 21 and 19 years old, respectively. This is considered to be related to women entering puberty earlier, as the onset of the disease is closely associated with hair growth, and puberty is a period of vigorous hair growth. However, in terms of incidence rate, the male-to-female ratio is 4:1 [7]. This phenomenon is believed to be due to the fact that during adolescence, women may pay more attention to personal hygiene and reduce mechanical friction caused by clothing compared to men. Meanwhile, men have an increased risk of external stimulation to the sacrococcygeal region due to their occupations, sports, and military service [8].

Devin R. Halleran [10] investigated postoperative recurrence factors in 307 patients. Statistical analysis showed that the postoperative recurrence rates for men and women were 33.3% and 33.1% respectively, with a P value of 0.92. Therefore, there is no statistically significant relationship between gender and recurrence of pilonidal sinus. In clinical practice, gender does not need to be a key factor in preventing postoperative recurrence [9].

4.2 Relationship between Age and Recurrence of Pilonidal Sinus

Pilonidal sinus is more common in young and middle-aged men, typically in those aged 17 to 25, mostly during adolescence. This is believed to be because during adolescence, the activation of hair follicle sebaceous glands increases, entering a period of vigorous sebum secretion [11]. During this period, hair growth increases, and adolescents are lively and active, creating a humid environment in the sacrococcygeal region, which increases the risk of pilonidal sinus. Although pilonidal sinus tends to occur during adolescence, recent studies have found that patients over 23 years old have a better prognosis, while younger patients have a higher probability of having a family history and a higher recurrence rate [12].

4.3 Relationship between Obesity and Recurrence of Pilonidal Sinus

Hair penetration is a key issue in the pathogenesis of pilonidal sinus. The gluteal region of obese people is humid and the skin is fragile, making them more prone to hair penetration. There is currently some controversy regarding the impact of obesity on recurrence, but it is clear that obesity is a contributing factor to the occurrence of pilonidal sinus.

4.4 Relationship between Hair and Recurrence of Pilonidal Sinus

As Karyadaki's theory indicates, hair is one of the important factors leading to pilonidal sinus. Postoperative hair removal in the gluteal cleft has a certain effect in preventing recurrence. Currently, common hair removal methods include shaving, cream hair removal, and laser hair removal, but their effects on postoperative recurrence vary. Studies have found that laser hair removal can reduce the postoperative recurrence rate of pilonidal sinus. Laser hair removal achieves permanent hair removal by destroying hair follicles, while shaving and cream hair removal require repeated operations and depend on patient compliance. As postoperative recovery progresses, patient compliance may decline, increasing the chance of hair falling into unhealed wounds and leading to short-term or long-term recurrence.

4.5 Relationship between Smoking and Recurrence of Pilonidal Sinus

It has been proven that smokers have poor wound healing after various surgeries. The mechanism is that smoking

impairs skin perfusion, leading to hair loss and premature skin aging. Among smokers with wound healing disorders, the proliferation rates of fibroblasts and macrophages are decreased, and growth factors are relatively deficient. In addition, nicotine can activate certain clotting factors and lead to the formation of microthrombi. Therefore, smoking can affect hair follicle health, cause hair disorders, and hinder postoperative wound healing. A study involving 610 patients who underwent surgery found that among smokers, the postoperative recurrence rate was significantly higher (1-year recurrence rate: 36% vs. 21%, P=0.008) [13]. Moreover, smokers had more wound complications than non-smokers (29% vs. 10%, P=0.005).

4.6 Relationship between Treatment Methods and Recurrence of Pilonidal Sinus

The treatment of pilonidal sinus mainly includes non-surgical (conservative) treatment and surgical treatment. Non-surgical treatment includes hair shaving or laser hair removal, antibiotic therapy, phenol injection, fibrin glue treatment, platelet-rich plasma treatment, and traditional Chinese medicine treatment. Surgical treatment mainly includes incision and drainage, primary suture after lesion resection, secondary healing after lesion resection, and minimally invasive treatment.

5. Non-surgical Treatment

5.1 Antibiotic Treatment

In the acute stage, the formation of pilonidal sinuses is usually accompanied by the formation of abscesses. Studies have found that patients with primary pilonidal sinus are prone to concurrent infections, mainly with anaerobic bacteria and Gram-negative bacteria [14]. Recurrent patients are mainly infected with aerobic/facultative anaerobic bacteria. Targeted use of antibiotics is meaningful for delaying the progression of pilonidal sinus, but no statistical significance has been found for the recurrence of pilonidal sinus so far [15].

5.2 Phenol Injection Therapy

Phenol injection therapy is one of the currently popular conservative treatment methods for pilonidal sinus. There are also literature studies on the impact of phenol concentration on the success rate of treating pilonidal sinus. A prospective randomized controlled study compared the effects of 80% and 30% phenol concentrations and found that after one-year follow-up, the recurrence rate of the former was lower, and the difference was statistically significant. However, there were no statistically significant differences in the healing time, the time to return to work, and the incidence of complications [16].

5.3 Fibrin Glue Therapy

It has been reported that patients are highly satisfied with the treatment of pilonidal sinus with fibrin glue, and most patients can return to normal life within one week of treatment. In terms of postoperative adjuvant therapy, Altinli et al. applied fibrin glue to patients after Limberg flap surgery and found that the wound drainage volume was less, the hospital stay was shorter, and all wounds healed [17]. No recurrence cases were observed during the 8-month follow-up.

5.4 Platelet-rich Plasma Therapy

It was reported that the success rate of PRP alone in the treatment of pilonidal sinus reached 97.1% after the first month, with an average healing time of 14 days, a median follow-up of 60.2 months, and a recurrence rate of 8.2% [18]. However, it is slightly regrettable that this method requires relatively high equipment standards and technical proficiency to be carried out.

6. Different Surgical Methods for Treating Pilonidal Sinus

Surgical treatment mainly includes incision and drainage, primary suture after lesion resection, secondary healing after lesion resection, and minimally invasive treatment, etc. Primary suture after lesion resection can be further classified into partial primary suture, transverse suture, midline suture and partial midline suture. Midline suture mainly refers to various flap surgeries, including Karydakis flaps, V-Y flaps, Z-plasty and other techniques as well as the improvements based on these techniques. Secondary healing after lesion resection is divided into open healing and bag suture. Minimally invasive treatments include curettage, laser therapy, endoscopic adjuvant therapy and radiofrequency ablation, etc.

6.1 Incision and Drainage

Pilonidal sinus, as an infectious disease, is prone to secondary acute infection accompanied by abscess formation. Incision and drainage is achieved by simply opening the abscess septum to ensure continuous and unobstructed drainage [19]. In 2022, an early observation study by American scholar Nixon on simple incision and drainage indicated that 58% of patients with incision and drainage healed within 10 weeks earlier, and 21% relapsed [20].

In 2014, Chesley [21] et al. studied that the recurrence rate of hairline incision and drainage was as high as 40%. With the continuous advancement of medical technology, in recent years, 90% of patients have healed within one month after incision and drainage surgery, and 10% to 15% of patients will relapse after healing. Incision and drainage do not address the cause of pilonidal sinus formation and are often accompanied by a high postoperative recurrence rate and chronic sinus tract formation.

6.2 Resection of the Lesion, Primary Suture and Secondary Healing

After a clear diagnosis of chronic infectious pilonidal sinus, surgical resection or minimally invasive treatment should be performed. Surgical resection mainly includes primary suture of the removed lesion and secondary healing. Both of them first design incisions based on the lesion range, then completely remove the lesion in the subcutaneous tissue of the skin to the sacral fascia, with the aim of removing all infected tissues, and finally suture the wound with the first-stage suture technique. The secondary healing technique involves open

healing without incision treatment or simple bag-shaped suturing at the edge of the incision skin.

In a meta-analysis conducted by Italian scholars Milone et al. in 2018 [22], it was pointed out that the recurrence rates of midline suture, partial midline suture, and open healing after up to 5 years of follow-up were 16.8%, 10%, and 17.9% respectively. Compared with secondary healing, primary suture is more acceptable in terms of wound closure aesthetics.

In 2020, a meta-analysis of the postoperative recurrence rate of 3,661 patients by Bi Siwei et al [23]. pointed out that midline suture has a relatively high recurrence rate, and the recurrence rate increases with the increase of sample size, age, and follow-up time.

In 2020, Swiss scholar Pfammatter treated children with non-complex sacrococcygeal pilonidal sinus using transverse primary suture. The result showed that transverse primary suture also had a lower recurrence rate than open healing (12.5% vs 37.5%). Contrary to the above conclusion, in a retrospective study conducted by American scholars Abraham et al. in 2021 involving 133 adolescent patients, the recurrence rate of primary suture and secondary healing after lesion resection was not statistically significant. The recurrence rate of secondary healing wounds was even lower, and the recurrence rate and complication rate decreased with age [24].

In 2020, Bi Siwei et al. also pointed out in a meta-analysis of the postoperative conditions of 3,383 patients that the recurrence rate after open healing was lower than that after primary suture [25, 26].

6.3 Bascom Hip Groove Elevation Surgery

In 1980, Bascom first described this technique, which involves making an oval incision to remove the skin and lesions, and freeing the contralateral flap to flatten the gluteal groove [28].

In 2020, British scholars Karim et al. randomly divided 60 patients with sacrococcygeal pilonidal sinus into the Bascom surgical group and the one-stage suture group for comparison. The results showed that the recurrence rate in the Bascom group was lower (3.3% vs 16.7%) [29].

A retrospective study by Danish scholars Syarre et al. on the 8-year follow-up after Bascom surgery in 2022 indicated that the cumulative recurrence rate over the past 10 years was 27% [27].

6.4 Limberg Flap

Limberg first described this technology in 1946. In a randomized controlled experiment conducted by British scholars Ray et al. in 2020 [30], it was pointed out that the Limberg flap technique had more advantages than the Bascom technique, but it did not reach statistical significance. In 2021, Russian scholars Danilov et al. performed Limberg flap surgery and simple excision and suture surgery on 117 patients with pilonidal sinus in groups, and there was a significant difference in the recurrence rate (1.6% vs 10.9%) [31]. In 2022, Pakistani scholars Zubair et al. conducted an observational study on 46 patients with sacrococcygeal sinus surgery using Limberg flaps and found that Limberg flap surgery has the advantages of low recurrence rate and few complications [32].

6.5 Karydakis Flap

In 1973, this technology was first used by Karydakis. In a retrospective study of 112 patients who underwent Karydakis surgery in 2020 by Polish scholars Cakari et al., the results [33] showed that the postoperative recurrence rate was 3.6% with an average follow-up period of 28 months. In a study conducted by Spanish scholars Golet et al. in 2021 involving 61 adolescent patients with pilonidal sinus, the Karydakis technique was proven to have a lower recurrence rate (4% vs 28.6%) than secondary healing after lesion resection [34]. In the same year, a meta-analysis of 1,943 patients with sacrococcygeal pilonidal sinus randomized controlled studies showed that there was no significant difference between the Karydakis flap technique and the Limberg flap technique in terms of complications, failure rate of healing, and length of hospital stay. The Karydakis flap technique had a higher cosmetic satisfaction rate among patients [35].

6.6 Keystone Flap

The Keystone flap, also known as the keystone flap, was first described by Behan et al. in 2003 and applied to the treatment of skin cancer in various parts of the body. It was not until 2021 that Turkish scholars Calisir et al. first applied this technique to the treatment of the sacrococcygeal pilonidal sinus. In 2021, Calisir conducted a retrospective study to analyze the postoperative conditions of patients who underwent Keystone flap surgery and Limberg flap surgery. The results showed that Keystone surgery had the advantages of low wound dehiscence and necrosis rates, short operation time, short healing time, and short time to return to work. Moreover, there was no significant difference in the recurrence rate between the two within one year [36].

6.7 Elliptical Rotating Flap

This technique was first reported in 2004 by Turkish scholars Nessar et al [37]. in the literature of the sacrococcygeal pilonidal sinus. The 20 patients were followed up for an average of 42 months after the operation and no recurrence was observed. Although the recurrence rate is 0%, this might be related to the sample size of this study.

In a recent study, Gundogdu et al. further modified this technique. The flap prepared was composed of not only the skin but also subcutaneous fascia, and its width was the same as the widest distance of the excised flap. This is called the subcutaneous fascia elliptical rotational flap. The study reviewed the relevant data of 149 patients who underwent surgery with this technique and pointed out that this technique has an effect similar to that of the elliptical rotating flap and its modified techniques. No recurrence was observed during the average 19-month follow-up [38].

At present, there are relatively few studies on elliptical

rotating flaps, and more comprehensive prospective studies are needed for verification.

6.8 V-Y Flap

This technique was first described in 1920 and has been successfully used as a triangular island flap. The main surgical operation is to first mark the lesion range and design a V-shaped flap. After the lesion is completely removed, the flap is freed. The flap is pushed towards the midline and then sutured. Finally, it is similar to a Y-shaped flap [39].

In 2017, Turkish scholars Akyuz et al. pointed out through a retrospective study that there was no statistical significance between the V-Y flap technique and the Limberg flap technique in terms of postoperative complications and recurrence rate, and the former had a longer hospital stay [40].

In a retrospective study conducted by Turkish scholars Koca et al. in 2018 on 61 patients with complex sacrococcygeal pilonidal sinuses (lesions ≥10cm or ≥1 sinus tract), the results showed that this technique had a relatively low incidence of complications. No recurrence was observed during the early or late follow-up after the operation [41].

In recent years, reports on V-Y flap technology have been very rare, and there is a great need for more scholars to conduct further in-depth research.

6.9 Z-plasty

As early as the 1860s, Z-plasty was applied to treat the sacrococcygeal sinus. The general process is as follows: First, a full-thickness longitudinal spindle-shaped resection of the lesion is performed at the center of the sacrococcygeal pilonidal sinus. Then, a Z-shaped transfer flap is designed. The two free flaps are both rotated in opposite directions to cover the defect area. Finally, they are sutured to present a "Z" letter shape. This technology can rotate the long axis of tissues, change the direction of scars and increase the length of skin in a certain direction. In 2020, Yang Yongping et al., in a retrospective randomized clinical study, pointed out that although Z-plasty has a longer operation time compared with simple resection, it has the advantages of fewer complications, shorter hospital stay and lower recurrence rate during short-term follow-up. Regrettably, there are currently few reports on the latest progress of this technology [42, 43].

7. Summary

There is still no definite conclusion on the best surgical approach for treating the sacrococcygeal pilonidal sinus, and it is highly controversial. The recurrence rates of different surgical procedures or the same surgical procedure in different geographical locations also vary, and further research is still needed at present.

Midline suture techniques (various flap techniques), secondary healing techniques, and minimally invasive treatments each have their own advantages and disadvantages. In current clinical studies, it is still undetermined which of the three has a better recurrence rate, but most are within an acceptable range.

Minimally invasive treatment has a shorter hospital stay and can quickly address the negative impact of the disease on the quality of life. Therefore, laser-assisted closure, VAAPS and Gips ring drill resection can be their preferred minimally invasive treatment methods.

It is recommended that the choice be made after a comprehensive assessment based on the patient's condition, wishes, and the surgical methods the surgeon is proficient in. For large lesions that are difficult to achieve with minimally invasive techniques, the currently widely used Bascom gluteal sulcus elevation, Limberg flap technique and Karydakis flap technique can be regarded as the preferred treatment methods.

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