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Review Article

PILONIDAL DISEASE

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Abstract. *Pilonidal disease is a common and well-recognized medical condition. It affects people in reproductive age, especially men and in combination with in-patient and outpatient treatment and absence from work it causes a considerable socioeconomic loss. This fact led to a renewed interest in understanding of the disease and search for the ideal method of treatment. The purpose of this review was to provide update on therapeutic options for patients with pilonidal disease. In case of chronic or recurrent pilonidal disease various treatment options exist, addressing different measures of surgical outcome. Like for many conditions, there is increase in the use of minimally invasive techniques in the treatment, which could be alternative to surgical excisions for pilonidal disease. Procedures for treatment of pilonidal disease can be divided in two large groups: minimally invasive treatment and excisional procedures. Although various treatment options exist nowadays, surgery is still preferred as definitive treatment. The optimal closure of the wound following an excision is still under debate since outcome measures depend mostly on type of closure selected. Most of the procedures fail to achieve the goals altogether. The final decision on treatment should be made based on surgeon and the patient' preference.*

Key words: *Pilonidal disease, pilonidal sinus excision, pilonidal surgery*.

Introduction

Pilonidal disease is a common and well-recognized medical condition. Its first description dates back to 1833, when Herbert Mayo described a sinus containing hair. In 1880, Hodge suggested the term "pilonidal", from the Latin word pilus, which means hair and nidus for nest (Fig. 1). Pilonidal disease (PD) affects people in reproductive age, especially men, which is in combination with in-patient and outpatient treatment and absence from work a considerable socioeconomic loss. This fact led to a renewed interest in understanding the disease and search for an ideal method of treatment [1].

The purpose of this review is to provide update on therapeutic options for patients with PD. Authors searched Medline using PubMed for articles in English language not older than ten years using search terms "pilonidal disease", "pilonidal sinus", "excision of pilonidal disease", "pilonidal disease guidelines". Older publications were hand searched and selected if considered relevant for the subject.

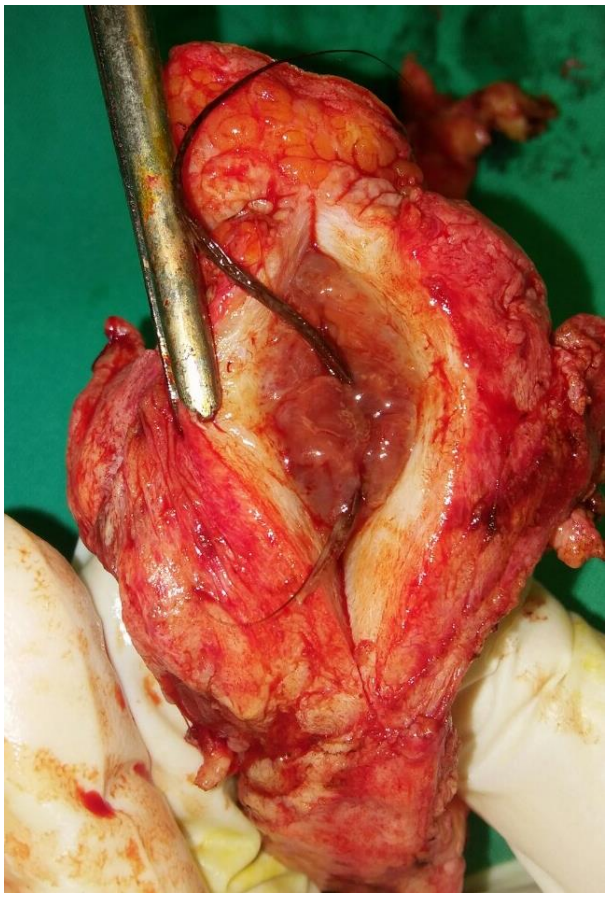

Fig. 1 Nest of piles (pilonidal disease)

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Incidence and risk factors

Incidence of pilonidal disease differs between continents. It's rare in Asia and Africa and high in Caucasians, highest recorded in Mediterranean area. Its incidence is rising due to reasons that are yet to be established. For example in Germany in 2012 there was increase from 29 patients per 100.000 earlier recorded to 48 per 100.000. It is probably due to sedentary life style, since even in population with small incidence rate, like Japan, pilonidal disease is seen more often in people with jobs requiring long sitting hours [2,3]

Pilonidal disease is more common in dark-skinned and dark-haired persons with excess body hair. Obesity is also recorded as risk factor [3,4]. Apart from these, male gender, poor body hygiene and excessive sweating are also described as risk factors. In the lack of prospective studies Harlak et al. published a prospective case control study in 2010 conducted among 587 patients and 2,780 healthy control subjects investigating risk factors for development of PD. According to their findings there are three most predictive risk factors: body hair rate, bathing habbits and sitting time. Using logistic regression analysis there was 219-fold higher risk for hairy people who shower or bathe two or less times per week and sit more than six hours per day compared with hairless people who shower or bathe three or more times per week and sit less than six hours a day to develop disease. The adjusted risk of PD was 6.33-fold greater for those who bathe two or less times per week than the risk for those people who take three or more baths per week, while the adjusted risk was 4.3-fold higher for individuals who were sitting more than six hours in a day. BMI was found to be a less important risk factor, but results might be different in investigation of community-based population, since this study included only active soldiers [5]. Later study also found irregular bathing as a risk factor for PD [4].

From its first description to the middle of 20th century PD is thought to be congenital. Probably the most cited theory of PD development is one by Gorge. E. Karydakis from General Army Hospital in Greece, who published his work in Lancet in 1973, after he examined 4670 previously operated Army candidates (with nearly 50% recurrence rate), and operated on 1687 patients using his new method [6]. He developed further the theory on hair insertions, which was first mentioned by Patey and Scarff in 1946. According to Karydakis PD is acquired and causative process that can be defined precisely using equation. The equation consists of three main factors which play a role in the hair insertion process: Hair (H) x Force (F) x Vulnerability (V). It can be used to calculate the possibility of PD. If these three main factors occur, then hair insertion and pilonidal sinus result. It is possible to list many secondary factors which together make up the three main factors, such as for example for H: number of loose hair, type or shape of hare, or for F: depth of natal cleft and friction; for V: wide pores, presence of wounds or scars at the natal cleft. All these factors not only explain all the known variations of the incidence PD, and the variation of its incidence in the same population over time, but also provide an answer to presence or absence of disease in some cases, for example, the presence of pilonidal sinus in some 'hairless' individuals, and its absence in others with hirsutism [7].

Experimental case matched study by Doll et al., compared mechanical strength of hair in occipital, lumbar, and intergluteal region and its relation to development of PD. The study has shown that vertical strength of occipital, lumbar, and intergluteal hair (along dorsal crest) from patients suffering from pilonidal disease was significantly greater than hair from their matched pairs. Cut hair fragments from occipital region were found in pilonidal nest which suggests that disease is related to this particular region as source. In concordance to their study authors are suggesting reduction in production of hair fragments in occipital (for example by shaving), removing cut hair along dorsal crest, reducing contact with hair within intergluteal fold such as with promptly showers after a hair cut in persons at risk [8].

Diagnosis and management

The diagnosis of PD is mostly established based on patient's history and clinical findings. Clinical finding are almost always visible characteristic pits in the intergluteal cleft, sometimes with hair extruding from their openings (Fig. 2). In recurrent disease or in chronic phase, sinus tract opening is visible (Fig. 3). According to Task force of the American Society of Colon and Rectal Surgeons from 2013, presacral mass should be

Fig. 2 Pits in intergluteal cleft

Fig. 3 Recurrent pilonidal disease

ruled out by digitorectal examination. Laboratory or imaging methods are not routinely used. It is important to distinguish PD from other conditions, such as perianal fistula, Crohn' disease or some infectious diseases (TBC, Syphilis or actinomycosis) [9]. Pilonidal disease can sometimes be mistaken for solitary hidradenitis suppurativa but unlike PD, hidradenitis affects more women than man. Their sonographic characteristics are also similar and accordingly PD might be a variant of localized form of hidradenitis suppurativa. Their histology is somewhat different, although histology finding cannot be always helpful in distinguishing PD from hidradenitis suppurativa [10].

Pilonidal disease can initially present as sacrococcygeal abscess. It is widely accepted that in this case incision and drainage are preferred method of treatment. Disease recurrence following this episode is reported to be 20% after 20 years, and there is no justification for wide excisions in this case since 80% of patients will be over treated and with substantial morbidity and doubled average time to return to work [11]. According to retrospective study from Australia on 134 patients with lateral longitudinal incision and 74 with midline incision, abscess with incision away from midline healed faster (p=0.02). Although admitting limitations of the study they concluded that pilonidal abscess should be drained away from the midline [12].

In case of chronic or recurrent disease various treatment options exist, addressing different measures of surgical outcome.

Like for many conditions there is increase in the use of minimally-invasive techniques in the treatment of PD, which could be alternative to surgical excisions for PD. Nowadays the treatment options could be divided in two large groups: minimally-invasive treatment or excisional procedures.

Minimally-invasive treatment of PD

Pit picking procedure was first described in 1965 by Lord and Millar who excised in local anesthesia affected pits using small elliptical excision, removed the hair and showed resolution in 32 out of 33 patients. Bascom was also one of the surgeons who recognized the importance of pit excision with addition of lateral incision for debridement of the sinus cavity [13]. In a recent study with adolescent patients, disease resolved in 92% of cases using this technique, although follow up period was limited to 5 months. According to authors advantages of this procedure is ease of performance in outpatient setting, it is well tolerated, requires minimal postoperative care and offers rapid recovery [14]. Reported recurrence rates with this procedure range from 10–20% [15].

Fibrin glue is often used in surgical practice. Its use in treatment of PD started in 2000. In 2018, results from single center study were published on 146 patients with PD of which 13% previously already had some kind of treatment. Procedures were done as one day surgery under general anesthesia (apart from one patient). After insertion of blunt probe for tract identification, sinuses were curetted and then flush with saline solution. Afterward the fibrin glue was inserted. No additional dressings were needed. Median operating time was 9 min. There were 27% of recurrences after the first glue application. Twenty four patients with recurrence decided for repeated treatment. Cumulative healing after 2 rounds was 96.9%. This procedure does not require technical equipment and can be easily thought and performed [16]. According to Cohran review current evidence is uncertain regarding the benefits associated with fibrin glue as monotherapy in PD or as adjunct to surgery. There is small number of low quality trials on the subject. RCT are needed to enroll larger number of patients measuring clinically relevant outcomes.

Phenol is also used in minimally- invasive treatment for many decades. It is a necrotizing material which causes burns on mucosa and skin. The preferred phenol is liquid (pure or 80%) or crystallized which turns into liquid form quickly at body temperature. Application tool is different according to the entrance technique. In the incision techniques, usually cotton swab with phenol is moved in the cavity. In the techniques without incision, one of the sinus openings are cannulated with a venous catheter or a blunt-ended metal trocar and phenol is injected into the sinus without pressure and left for 1–3 minutes. The injected volume of phenol is mean 1.7±1.9 ml. The sinus is then washed out with normal saline to prevent phenol leakage. The patients treated under local anesthesia are able to leave the hospital immediately after the procedure. Recurrence is regarded as occurrence of the same complaints after asymptomatic period and a second cause of treatment failure. Most studies mentioned that repeated applications were done if necessary (continuation of purulent discharge). Time interval between the repeated applications was variable among the studies (from 1–6 weeks). Wound healed long but there was immediate return to activities. Satis-

factory results with no evidence of recurrence or prolonged discharge were obtained in 60–100% of the patients [18]. Up to date there is only one RCT trail that compared effect of phenol injection with excision and open healing. Study included 140 patients equally distributed in both arms. Time to complete wound healing (16.2 ± 8.7 versus 40.1 ± 9.7 days) was significantly in favor of the phenol injection group ($p<0.001$). The median operation time was significantly shorter ($p< 0.001$). Pain score after surgery as well as painkiller intake were also in favor of phenol group. At the mean follow-up of 39.2 ± 9.0 months no difference was seen in the recurrence rate between the two arms. Authors of the study concluded that phenol injection is as effective as the excision with open healing in the PD treatment [19]. Phenol therapy can be combined with other methods of treatment such as video-assisted diathermy ablation of the sinus cavity, with achievement of good results after 22 months of follow up in terms of fast wound healing and low recurrence [20].

Endoscopic treatment of PD implies ablation of sinus tract using video assisted guidance. In the literature it is found under the name of EPSiT (endoscopic pilonidal sinus treatment) or VAAPS (video-assisted ablation of pilonidal sinus). View through the endoscope allows identification of lateral tracts [21]. A recent systematic review and meta-analysis of endoscopic treatment of PD with nine studies and 497 patients was published. The mean operating time was 34.7 ± 17.7 min. Procedure was performed as day-case surgery in all. Seven studies reported the pain VAS (measured from 0–10) within the first week after the procedure, the mean VAS was 1.35 ± 0.8 (range, 0.5–2), while 36 (8.6%) patients required intravenous analgesics in the first postoperative day. Failure of the technique was recorded in 40 (8.04%) patients, 20 (4.02%) had persistent (non-healing) pilonidal sinus, and 20 (4.02%) developed recurrence of SPD after complete initial healing of the primary wound. The weighted mean failure rate of the technique was 6.3% (95% CI 3.6–9.1). Failure of the technique was managed with redo of endoscopic treatment in 24 patients. Complication rate across the study ranged between 0 and 11.1%. Complications included hematoma, infection, persistent discharge, and failure of healing. The mean weighted complication rate was 1.1% (95% CI 0.3–2.4). The mean time to complete healing after the procedure was 32.9 ± 23 days. The mean time to return to work and normal activities was 2.9 ± 1.8 day. Ninety five percent were completely satisfied with the procedure. Authors conclude that endoscopic treatment of PD is a novel and promising method whose main advantage over conventional surgery are mild postoperative pain, quick healing, and short time to return to work and daily activities. The long-term outcome of the procedure is still unclear and longer follow-up is needed [22].

After good initial results Georgiou published data on Pilonidal disease Laser Treatment (PiLaT) in patients with primary disease. Patients with disease recurrence were excluded. In local anesthesia and in prone position after debridement and flushing with saline solution of pits and sinus tracts, a 1–2 mm metallic probe was inserted into sinus. Energy was deliver trough a tip of the probe in circumferential manner in order to shrink and obliterate tract. Primary end point of the study was healing at 8 weeks, and preservation of these results up to 12 months. After one year out of 60 patients enrolled, overall success rate was 92%. All of patients who failed the first time except for one, agreed to undergo again PiLaT procedure. With this results success rate reached 98% [23]. Similar results were earlier published in Belgium with reported recurrence of 2.9% and success rate of 87.5% [24]. Both studies conclude that it is safe, highly effective, almost painless and easy to learn and to perform and should be offered to all patients. Drawback of this procedure is its cost (around 600 euro), which could be balanced with earlier return to work. These studies included small number of patients (60 and 40), and promising results should be evaluated through longer follow up and in RCT.

Excisional procedures

Open excision is the approach most frequently implemented globally. Complete resection of the sinus is followed by thorough curettage of the cavity. The wound is left to heal by secondary intention or wound edges can be marsupialized. The disadvantage of this procedure is long healing time with delayed return to work. Reported recurrence rates vary greatly from 2% to 35% [25]. Meta-analysis on 26 RCT and 2530 patients compared open wound to primary closure. Wound with primary closure did heal faster. On the other hand recurrence rates were lower for open wounds (RR 0.60, 95% CI 0.42 to 0.87). This meta-analysis did not show difference in rates of SSI between two groups [26]. Randomized control trial compared Limberg flap procedure to secondary wound healing following excision (Fig. 4). Limber flap procedure took longer 60 (30–80) vs. 30 (10–75) minutes ($p < 0.001$) and had higher

Fig. 4 Limberg flap procedure

complication rate (49% vs. 12%, $p < 0.001$). Limberg flap procedure failed to show advantage over open wound mainly due to high complication rate [27]. One of the conclusions of the previously mentioned meta-analysis was that there was advantage of off-midline over midline wound closure, although in some studies midline suturing is of no importance if it is done in tension free manner [26]. Sevinc et al., conducted a RCT comparing midline and off-midline closure. Patients with primary PD were randomly assigned into 3 groups (two different flap procedures and tension free primary closure), 50 each. Main outcome measures were complications and recurrence. The groups were similar in terms of infection rate and development of seroma. The mean painless sitting time was significantly shorter in primary group. The median follow up time of the study was 24.2 months and the recurrence rates were similar (p=0.876). According to the authors of the study release of subcutaneous tissue enabling tension free suture line eased wound healing [28].

Off-midline procedures, such as Karydakis flap or Limberg rotation flap are oriented towards flattening of the natal cleft. Low recurrence rate after originally described Karydakis procedure, is due to simple objective "no raphe, no wound and scar at the depth". The intact skin put at the depth seems not to inherit the vulnerability of the raphe. The natural depth of the intergluteal fold, the raphe, is invaded by the hair and scar in the midline can easily become new entry point [6].

Meta-analysis from 2018 tried to give an answer to the question, which off-midline procedure is most appropriate. Eight studies involving 1121 patients were included. Patients were operated with either Karydakis flap (KF) or using Limber flap reconstruction. All of the studies were conducted between year 2004 and 2013. Long time follow up rate ranged from mean 15.5 to 33.3 months. In some studies modified Limberg flap technique was used, by performing excision with the lower border of the rhomboid 2 cm lateral from to the cleft. In subgroup analysis there was no difference between modified Limberg flap (LF) and conventional LF in all outcome measures including recurrence, except for postoperative rate of seroma. No statistically significant difference in recurrence rate between LF and KF was noted (OR=1.07; 95%CI[0.59-1.92]; p=0.83. There was significant difference in terms of seroma favoring LF [OR=2.03; 95%CI[1.15-3.95]; p=0.01). There were no differences in wound rupture nor in wound infection rate. Data on overall morbidity could not be pooled due to high statistical heterogeneity. In two studies statistically significant difference were found favoring LF, while in remaining six morbidity was comparable. KF required shorter operating time in all studies. Most of the studies report similar time to return to work in both arms [29].

The recurrence rate is the most important variable for the comparative assessment of different treatment modalities, although in most publications recurrence is not defined. In some cases surgical treatment is followed by a non-healing wound that sometimes requires second operation. From the academic perspective, absence of wound healing is not correctly designated as a recurrence, but from the patient's point of view, the only relevant fact is that repeat surgery must be done. For that reason the term "treatment failure" would be preferable [15].

When it comes to long term patient satisfaction recurrence is most important. Study on 583 male patients from military cohort with long follow up (7-22 years) investigated patient satisfaction with surgical treatment in terms of in-hospital time, outpatient treatment, pain, aesthetic impression and long term recurrence rate. According to the results patients are dissatisfied with the results of any surgical technique, if they experience recurrent disease. Pain during wound treatment and cosmesis has no influence on patient satisfaction in the long term, as long as they are recurrence-free over the next 20 years [2].

Recurrence rate following surgery for PD depend on follow-up time. This was pointed out in the meta-analysis and merge analysis from 2018, on combined RCT/non-RCT studies with 89,583 patients available from 1833 to 2017. This dependence, i.e. the steepness of increase of recurrence with longer follow-up times, is specific to a surgical procedure. According to these data primary midline closure should be abandoned, while older therapies (such as marsupialization) may be reconsidered. Flap procedures (Karydakis, Bascom, Limberg) and asymmetric procedures are superior, as proven by RCT and combined RCT/non-RCT trials. Follow-up of PSD patients should always be planned long term, i.e., five or ten years for reliable conclusion [30].

Currently there are three guidelines for the treatment of PD available, all made by relevant surgical societies, from Italy, USA and Germany [31-33]. In Germany despite the increasing number of novel procedures most surgeons still prefer traditional methods. The level of evidence on most topics is moderate or low, while most of RCT are lacking a power calculation and do not describe allocation concealing. Most trials have been performed in Middle East and Southern Europe, which questions the applicability of the obtained results [31]. Patient needs to be adequately informed about all aspects of treatment, from possible complications, cosmetic effects, postoperative course, to risk for recurrence [32]. Clinical Practice Guidelines Committee of the American Society of Colon and Rectal Surgeons published the latest guideline for PD treatment in 2019 [33]. Based on low quality evidence there is weak recommendation for elimination of hair from the intergluteal cleft and surroundings in both, acute and chronic pilonidal disease, in the absence of abscess as a primary or adjunct treatment measure. Based on moderate quality evidence in patients with acute or chronic pilonidal disease without abscess, phenol application is an effective treatment that may result in rapid and durable healing (strong recommendation). Fibrin glue may be effective as a primary or adjunctive treatment of PD (weak recommendation based on moderate-quality evidence). Patients who require surgery for chronic PD may undergo excision and primary repair (with consideration for off-midline closure), excision with healing by secondary intention, or excision with marsupialization. Flap-based procedures may be performed, especially in the setting of complex and recurrent chronic PD when other techniques have failed (strong recommendation

based on moderate-quality evidence). Minimally invasive approaches to acute and chronic PD that use endoscopic or video assistance may be used but require specialized equipment and expertise (weak recommendation based on moderate-quality evidence). It is emphasized, that these guidelines should not be inclusive of all proper methods of care nor exclusive of other methods [33]. The final decision on treatment has to be individualized for every patient taking into consideration also surgeon experience and confidence in performing different techniques [32,33].

Conclusion

Although various treatment options exist nowadays surgery is still preferred as definitive treatment. The optimal closure of the wound following an excision is still under debate since outcome measures depend mostly on type of closure selected. Most of the procedures fail to achieve the goals altogether. The final decision should be made based on surgeon and patients' preference.

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