

Pilonidal disease specimens of 905 patients revealed no malignancy, however we still insist on histopathological examination

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Abstract

Aim: Pilonidal disease presents with acute abscess, sinuses, pits and scarring in the sacrococcygeal region. Surgical intervention is one of the treatment options of pilonidal disease. However, it has been a matter of debate, whether histological analysis of surgically resected pilonidal disease specimens would be necessary.

Material and Methods: Medical records of the patients with pilonidal disease who underwent either primary excision and closure or modified Limberg flap surgery were reviewed retrospectively. The results of the histopathological examination of the pilonidal disease specimens were evaluated.

Results: This study included 905 patients (686 male and 219 female) with pilonidal disease who were treated with surgical intervention. The mean age of the patients was 23.96±7.5 years. All surgically excised pilonidal disease specimens of the patients were histopathologically evaluated in order to rule out malignancy. No malignancy was detected.

Conclusion: We conducted a Pubmed search with the term "pilonidal" between January 2000 and March 2019. We identified 91 patients with malignant degeneration arising on pilonidal disease in the English medical literature. The most common pilonidal disease associated malignancy was squamous cell carcinoma. Even it is rare, malignancy may accompany pilonidal disease. Therefore, even we have not encountered any malignancy in 905 patients with pilonidal disease, we still insist on histopathological examination of all surgically resected pilonidal disease specimens in order to rule out malignancy.

Keywords: Histopathological examination; malignancy; pilonidal disease; squamous cell carcinoma

INTRODUCTION

Follicular occlusion tetrad consists of pilonidal disease, hidradenitis suppurativa, acne conglobata and dissecting cellulitis, which share similar pathophysiology. Follicular occlusion in the apocrine glands is thought to be the common triggering factor in the development of these conditions (1).

Pilonidal disease presents with acute abscess, chronic sinuses with intermittent drainage, pits and scarring in the sacrococcygeal region (2). Pilonidal disease affects 0-5% of the general population (3). It is more common in men than in women. The patients are usually between the ages of 15 to 30 (2). It has been suggested that obstruction of the infundibulum of the hair follicle as a result of follicular hyperkeratosis leads to dilated and ruptured hair follicles, secondary infection, fistulae and abscess (4). Loose hairs

in the gluteal cleft which penetrate the skin have also been implicated in the etiopathogenesis of pilonidal disease. Foreign body reaction caused by penetrated hair follicles may cause the formation of midline pits and secondary infection (5). The diagnosis of pilonidal disease is usually made based on patient history and clinical examination. Midline pits in the gluteal cleft are the characteristic physical findings. However, differential diagnosis should also include hidradenitis suppurativa, furuncle, Crohn's disease, perianal fistula and infections like tuberculosis, syphilis, and actinomycosis (5).

Moreover, the clinician should be aware that long-lasting pilonidal disease can result in malignant transformation. The malignancy type arising from pilonidal disease is usually squamous cell carcinoma. However, basal cell carcinoma, sweat gland adenocarcinoma and verrucous

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carcinoma have also been reported to be associated with pilonidal disease (6). The incidence of squamous cell carcinoma arising from pilonidal disease is approximately 0.1%. Therefore, early treatment is recommended in sacrococcygeal fistula as chronic inflammation increases the risk of malignant degeneration (3). Hereby, we report the results of histopathological evaluation of 905 patients (686 male and 219 female) with pilonidal disease, who underwent surgery.

MATERIAL and METHODS

This study included patients between 12 and 66 years of age with pilonidal disease, who were admitted to the pediatric and adult general surgery outpatient clinics between January 2015 and March 2019. Local ethics committee approval was obtained (2019/03-15). The medical records of the patients with pilonidal disease

who underwent surgical intervention were reviewed retrospectively. The surgical technique was either primary excision and closure or modified Limberg flap surgery. All pilonidal disease surgery specimens were histopathologically evaluated. Exclusion criteria were having cancer, human papilloma virus infection of the anogenital area, organ transplantation, receiving immunosuppressive medications, radiotherapy, chemical exposure like arsenic, industrial tar and paraffin.

RESULTS

The study included 905 patients (686 male, 219 female) with pilonidal disease. The mean age of the patients was 23.96 ± 7.5 years (range: 12 - 66). The mean age of the males was 25.35 ± 7.57 years (range: 13 - 66). The mean age of the females was 19.62 ± 5.32 years (range: 12 - 45).

Table 1. Pilonidal disease and malignancy

Author	Age / Gender / Duration	Diagnosis
Delvecchio et al., 2019	83 / M / 30 years	SCC
Michalopoulos et al., 2017	60 / M / 7 years	SCC
Esposito et al., 2015	63 / M / 43 years	SCC
Parpoudi et al., 2014	77 / M / 10 years	SCC
Cruz-Mendoza et al., 2014	60 / M / 50 years	Epidermoid carcinoma
Eryilmaz et al., 2014	44 / M / 10 years	Low grade SCC
Pandey et al., 2014	58 / M / 19 years	Moderately differentiated SCC
Erdogan et al., 2013	63 / M / 35 years	SCC (Verrucous carcinoma)
Goyal et al., 2013	70 / M / 21 years	SCC
Nunes et al., 2013	61 / M / 10 years	Well differentiated SCC
De Martino et al., 2011	60 / M / 15 years	SCC
Alarcon-Del Agua et al., 2011	36 / M / 10 years	SCC
	57 / M / 12 years	SCC
	62 / M / 40 years	SCC
	62 / F / 1 year	BCC
Yetim et al., 2011	52 / M / 20 years	SCC
Bolandparvaz et al., 2009	52 / M / 6 years	Moderately differentiated SCC
Tirone et al., 2009	43 / M / 14 years	SCC
Chatzis et al., 2009	50 / M / 15 years	SCC
Sharma et al., 2009	52 / M / 20 years	Well differentiated SCC
Mentes et al., 2008	48 / M / 10 years	SCC (Verrucous carcinoma)
Kovacevic et al., 2007	57 / M / 17 years	Well differentiated SCC
	49 / M / 21 years	Poorly differentiated SCC
Malek et al., 2007	40 / F / 5 years	SCC
Frost et al., 2007	80 / F / 59 years	Well differentiated SCC
Agir et al., 2006	49 / M / 15 years	SCC
Alecha Gil et al., 2006	48 / M / 25 years	Epidermoid carcinoma
Adanali et al., 2002	55 / M / *	SCC
Cilingir et al., 2002	42 / M / 16 years	SCC
Atmatzidis et al., 2002	61 / M / 30 years	SCC
Pekmezci et al., 2001	56 / M / 36 years	Malignant degeneration
Velitchklov et al., 2001	49 / M / 22 years	SCC
De Bree et al., 2001	48 / M / 25 years	Malignant degeneration
	59 / M / 30 years	
	67 / M / 5 years	

SCC: Squamous cell carcinoma, BCC: Basal cell carcinoma, M: Male, F: Female, * This information could not be retrieved from the article.

De Bree et al. reported 56 patients with malignant degeneration arising on pilonidal disease until 2001 (35). In addition to that, we have identified 35 patients with pilonidal disease associated malignancy since 2001 to date (Table 1) (3,6,8,10-35,36). Thus, a total of 91 patients with pilonidal disease were diagnosed with malignancy. The most commonly detected tumor type was squamous cell carcinoma (SCC)

The histopathological evaluation revealed that none of the 905 pilonidal disease surgery specimens contained malignant degeneration.

DISCUSSION

Pilonidal disease presents with suppurative abscess or intermittent draining sinuses in the sacrococcygeal region. Pilonidal disease is usually treated with surgical excision. Histopathological examination of the pilonidal disease specimen usually reveals granulomatous pseudo-cystic formation of the subcutaneous tissue with hair debris. However, most of the surgeons prefer histopathological evaluation of the surgical specimen in order to rule out malignancy, which could arise on pilonidal disease (7).

Boulanger et al. reported that no malignancy was observed in the histopathological evaluation of surgical specimens of 731 patients with pilonidal disease. Therefore, Boulanger et al. questioned whether routine histopathological evaluation of all pilonidal disease surgery specimens was essential. Boulanger et al. recommended histopathological evaluation for patients older than 50 years, and for lesions either with atypical presentation or prolonged duration (7). Furthermore, excessive growth of the ulceration, groin lymphadenopathy and human papillomavirus infection have also been associated with malignancy (8,9).

Neoplastic changes have been reported in 0.1% of the histopathological evaluation of pilonidal disease lesions. The risk for malignancy development is especially increased in patients who had pilonidal disease longer than 20 years. The type of malignancy arising on pilonidal disease is mostly squamous cell carcinoma (SCC), which forms approximately the 88% of the patients. Local tumor invasion and spread to underlying bones, anus, rectum, or lymph nodes in the groin may be observed. However, SCC usually does not spread to other organs (3).

Patients who had malignancy associated pilonidal disease have been reported as case reports in the English medical literature. We have identified 35 patients who had pilonidal disease diagnosed with malignancy since 2001 to the date (Table 1) (3,6,8,10-36). In addition, De Bree et al. reported that 56 patients with malignant degeneration arising on pilonidal disease were described in the medical literature until 2001 (35). Therefore, a total of 91 patients were diagnosed with malignancy associated pilonidal disease. The most commonly detected tumor type was SCC. On the contrary, we did not observe any malignant transformation in the histopathological evaluation of the surgical specimens of 905 patients with pilonidal disease. Moreover, literature search revealed that malignant changes could also be encountered both in young patients and patients with short disease duration (19). In addition, tumor arising on pilonidal disease tends to be locally aggressive (3). Therefore, clinicians should be careful in the management of pilonidal disease. Even it is rare, malignancy may accompany pilonidal disease

Furthermore, there are other minimally invasive approaches available for the management of pilonidal disease such

as crystallized or liquid phenol application, fibrin glue administration and video-assisted ablation of pilonidal sinus tract. During these procedures neither a biopsy is taken nor is a pathological examination performed. This raises the question, if some of the malignant cases associated with pilonidal disease are missed after these procedures. Therefore, surgeons should also consider taking a biopsy during minimally invasive treatment of pilonidal disease.

CONCLUSION

As a result, even we have not encountered any malignancy in 905 patients with pilonidal disease, we still insist on histopathological examination of all surgically resected pilonidal disease specimens, in order to rule out malignancy.

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