Introduction

Pilonidal cyst or pilonidal sinus is a common inflammatory disease usually appearing in the sacrococcygeal area. Its incidence in the general population is 26/100000, while it affects mostly working males between the age of 15 to 30 (1, 2). Keratin plugs, foreign substance reaction due to hair, as well as dermopathy and debris in hair follicles in the natal cleft are considered the main causative factors, while not only the disease itself, but also its treatment cause discomfort and deterioration of the patient’s quality of life. The recurrence rates after surgical treatment have been reported to range from 0 to 4% (2, 3).

We present a case of 33-year old male, with a follow up of a whole year, who presented with recurrent pilonidal sinus due to an epidural abscess. A 33-year old male, suffering from previously undiagnosed and untreated diabetes mellitus (DM), presented to our emergency department (ER), one month after open surgical treatment of pilonidal sinus, due to weakness and fever. After re-operation of the pilonidal cyst and due to post-operative pus production and continuation of fever a computer tomography (CT) scan was performed revealing an epidural abscess extending from the thoracic vertebrae 12 (T-12) to the sacrococcygeal area. At that point he underwent new surgery for drainage of the epidural abscess. The patient received intravenous antimicrobial treatment and was discharged on the 23rd postoperative day without signs or symptoms of infection. At follow up for a whole year no signs of recurrence have been observed.

Case report

A 33-year old male, working in a waste company, with a Body Mass Index (BMI) of 32.8 presented to the Emergency Department (ER) due to weakness and fever, one month after open surgical treatment of pilonidal sinus under local anesthesia. Clinical examination revealed recurrence of the pilonidal cyst with pus production. The patient was febrile (38°C), alert and had stable vital parameters.

Initial laboratory investigation showed white blood cell (WBC) count = 9000/mm$^3$, c-reactive protein (CRP) = 154 mg/L, glucose (Glu) = 346, while pus, blood and urine cultures were obtained. The examination at the ER revealed that the patient had diabetes mellitus (DM), previously undiagnosed.

On the 3rd day of hospitalization he underwent, under local anesthesia, open surgical treatment for the recurrent pilonidal cyst with pus production. The patient was febrile (38°C), alert and had stable vital parameters.

Corresponding author: Christos Koutserimpas, e-mail: chrisiku91@hotmail.com
© Copyright 2017, CIC Edizioni Internazionali, Roma

SUMMARY: Epidural abscess imitating recurrent pilonidal sinus: a case report.

C. KOUTSERIMPAS1, N. NIKITAKIS1, A. SKARPAS1, M. LADA2, E. PAPACHRISTOU1, G. VELIMEZIS1

Pilonidal sinus or pilonidal cyst is a common benign disease, affecting mostly young working men. We present the first case of an epidural abscess imitating pilonidal sinus. A 33-year old male, suffering from previously undiagnosed and untreated diabetes mellitus (DM), presented to our emergency department (ER), one month after open surgical treatment of pilonidal sinus, due to weakness and fever. After re-operation of the pilonidal cyst and due to post-operative pus production and continuation of fever a computer tomography (CT) scan was performed revealing an epidural abscess extending from the thoracic vertebrae 12 (T-12) to the sacrococcygeal area. At that point he underwent new surgery for drainage of the epidural abscess. The patient received intravenous antimicrobial treatment and was discharged on the 23rd postoperative day without signs or symptoms of infection. At follow up for a whole year no signs of recurrence have been observed.

KEY WORDS: Pilonidal cyst - Pilonidal sinus - Epidural abscess - Pilonidal disease - Recurrence.
Postoperatively the production of pus from the open wound continued, while the patient remained febrile (38). On the 5\textsuperscript{th} postoperative day he had a computer tomography (CT) scan that revealed an epidural abscess expanding from the height of the 12\textsuperscript{th} thoracic vertebrae until the sacrococcygeal area (Figure 1).

At that point the patient was re-operated under general anesthesia. A total of 4 incisions were performed; two at the lower level of the lumbar spine and two at the gluteus maximum area and the abscess was drained, while after creating a tunnel between the open wound from the pilonidal sinus surgery and those 4 incisions, 4 additional drainages were placed (Figure 2). The i.v. antimicrobial treatment was continued.

A control CT and magnetic resonance imaging (MRI) were performed at the 10\textsuperscript{th} postoperative day, both showing a remaining abscess with no signs of spondylo-discitis (Figure 3). On the 15\textsuperscript{th} postoperative day a CT-guided pus-drainage was performed and the culture of the drained material revealed the same microorganism, while acid fast microscopy and subsequent culture for mycobacteria were negative.

Two days after the CT-guided drainage he became afebrile while his WBC reached 6000/mm\textsuperscript{3}. The patient had a satisfactory recovery and was finally discharged on the 23\textsuperscript{rd} postoperative day, receiving cefuroxime per os for 15 days and was followed up in the outpatient clinic. The two lower placed drainages had been removed 3 days before discharge, while the remaining two during the patient’s follow up (7 days after discharge).

At the out-patient clinic and after receiving a total of
15 days per os antimicrobial treatment, the patient had no complaints and the clinical examination did not reveal signs or symptoms of recurrence. He continued treatment and monitoring of his DM.

After a whole year of close follow-up the patient looks fine, while a new CT scan confirmed the clinical findings (Figure 4).

Discussion

Pilonidal disease is a very common, well recognized by clinical examination, benign disease, occurring more frequent in men (2, 4). We presented the first, to our knowledge, case of epidural abscess imitating recurrent pilonidal disease. After a comprehensive electronic search of the international literature, using the PubMed and Cochrane database, a link between pilonidal disease and epidural abscess appears only due to the use of epidural anesthesia (5). This does not apply to the present case, since our patient underwent surgery under local anesthesia.

There could be a couple of factors that led to this extremely rare clinical presentation of a recurrent pilonidal disease caused by an epidural abscess in a 33 year old male, such as the unsanitary conditions of his occupation and the previously undiagnosed DM.
It is of note that his occupation was a garbage worker in an unhealthy environment. Poor hygiene conditions dominated his work, as well as his personal life.

Additionally, he was suffering from DM. DM is considered one of the largest emerging threats to health in the 21st century. It is a clinical syndrome related to deficiency of either insulin secretion or action. Patients with DM have infections more frequently than the rest of the population. Furthermore, these infections are more complicated (6, 7).

The pathophysiology of how DM leads to infections is multifactorial (7). Some studies have shown a deficiency of the C4 component and less secretion of interleukin-1 (IL-1) and interleukin-6 (IL-6) in response to lipopolysaccharides (8, 9). Furthermore, due to the increased glycator-alpha (TNF-α) is inhibited (10). Hyperglycemia can also lead to impaired antibody function, increased apoptosis of polymorphonuclear leukocytes, as well as reduced polymorphonuclear leukocyte transmigration through the endothelium (7, 10). Additionally, a high glucose environment is friendlier to some microorganisms (6).

DM depresses the immunity and is a causative factor for a plethora of infections (6). A physician must keep a keen eye for infection sources in DM patients, not only due to the higher prevalence, but also due to the complications that can appear, such as hypoglycemia, ketaoadisis and coma.

Conclusions

Although common and generally benign, pilonidal disease could also hide some more serious conditions (11, 12). Squamous cell, basal cell, adeno- and verrucous carcinomas have also been reported as causative factor for pilonidal sinus (13-15).

In conclusion, it is important that a thorough clinical examination in patients with pilonidal cyst is performed and that every pilonidal sinus lesion should be sent for histopathologic examination. An epidural abscess as a causative factors for pilonidal sinus is extremely rare. However, it should be reported and kept in mind in recurrent cases.

Declarations

Acknowledgements: None.
Funding: None.
Conferences: None.
Authors’ contributions: CK, NN, AS and EP for the literature search and analysis, and manuscript writing. ML and GV for the final manuscript revision. All authors have read and approved the final manuscript.
Conflict of interest: The Authors declare that there is no conflict of interest.
Consent for publication: received.

References

1. McCallum IJ, King PM, Bruce J. (2008) Healing by primary closure versus open healing after surgery for pilonidal sinus: systematic review and meta-analysis. BMJ. 2008;336:868-871.
2. Farrell D, Murphy S. Negative pressure wound therapy for recurrent pilonidal disease: a review of the literature. J Wound Ostomy Continence Nurs. 2011;38:373-378.
3. Shafik A. Electrocauterization in the treatment of pilonidal sinus. Int. Surg. 1996;81:83-84.
4. Lee PJ, Raniga S, Bryani DK, Watson AJ, Faragher BG, Frizele FA. Sacrococcygeal pilonidal disease. Colorectal Dis. 2008;10:652-656.
5. Verdu A, Garcia-Granero E, Garcia-Fuster MJ, Martín A, Millán M, Lledo S. Lumbar esstomyelitis and epithelial abscess complicating recurrent pilonidal cyst: report of a case. Dis Colon Rectum. 2000;43:710-714.
6. Poleg AV, Weeratunga T, McCarthy JS, Davis TM. Common infections in diabetes: Pathogenesis, management and relationship to glycemic control. Diabetes Metab Res Rev. 2007;23:3-13.
7. Geerlings SE. Urinary tract infections in patients with diabetes mellitus: Epidemiology, pathogenesis and treatment. Int J Antimicrob Agents. 2008;31:554-7.
8. Geerlings SE, Hoepelman AI. Immune dysfunction in patients with diabetes mellitus (DM). FEMS Immunol Med Microbiol. 1999;26:256-65.
9. Geerlings SE, Brouwer EC, Van Kessel KC, Gaaster W, Stolk RP. Hoepelman AI. Cytokine secretion is impaired in women with diabetes mellitus. Eur J Clin Invest. 2000;30:995-1001.
10. Nirmal J, Capuro GM, Weitekamp MR, Karchmer AW. Infections in patients with diabetes mellitus. N Engl J Med. 1999;341:1906-12.
11. Humphries AE, Duncan JE. Evaluation and management of pilonidal disease. Surg Clin North Am. 2010;90:113-24.
12. Eryilmaz R, Sahin M, Olan I, Alimoğlu O, Somay A. Unusual pilonidal sinus disease: predisposing factors and treatment. World J Surg. 2005;29:1158-60.
13. Chatzis I, Ntousas G, Kounsuzaki A, Chatzitheoklitos E. Squamous cell carcinoma related to long standing pilonidal-disease. Eur J Dermatol. 2009;19:408-9.
14. Sharma D, Pratap A, Ghosh A, Shukla VK. Malignant transformation of a pilonidal sinus. Surgery. 2009:145:243-4.
15. Wolff H. Carcinoma auf dem Boden des Dermoids. Arch Klin Chir. 1900;62:731-8.