

Contents lists available at [ScienceDirect](http://www.sciencedirect.com)

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Intermammary pilonidal sinus: The first case series



Seerwan Hama Shareef^a, Tahir A. Hawrami^a, Abdulwahid M. Salih^a,
Fahmi H. Kakamad^{b,c,*}, Hawbash M. Rahim^c, Hunar A. Hassan^c, Dahat A. Hussein^c

^a Faculty of Medical Sciences, School of Medicine, Department Surgery, University of Sulaimani, François Mitterrand Street, Sulaimani, Kurdistan Region, Iraq

^b Faculty of Medical Sciences, School of Medicine, Department Cardiothoracic and Vascular Surgery, University of Sulaimani, François Mitterrand Street, Sulaimani, Kurdistan Region, Iraq

^c Kscien Organization for Scientific Research, Hamdi Street, Sulaimani, Kurdistan Region, Iraq

ARTICLE INFO

Article history:

Received 30 July 2017

Received in revised form 10 October 2017

Accepted 14 October 2017

Available online 20 October 2017

Keywords:

Pilonidal sinus
Intermammary
Excision

ABSTRACT

INTRODUCTION: Pilonidal sinus (PNS) is an inflammatory condition caused by hair penetration into the epidermis. It occurs usually in sacro-coccygeal area. Intermammary pilonidal sinus is a very rare variant with a few case reports in literature. The aim of this study is to highlight the presentation and management of intermammary pilonidal sinus.

METHOD: A retrospective multicenter case series. Including all cases of intermammary PNS admitted to either of 5 major tertiary hospitals in Kurdistan region of Iraq. Inclusion criteria were any case of intermammary PNS diagnosed by histopathological examination. The data were taken from the medical records and direct interview with the patients.

RESULTS: The case series included 12 patients, all case were female. The age range was from 13 to 29. All of cases were obese with body mass index (BMI) ranged between 30.1–32.2 kg/m². All of them presented with discharge for a long time ranging between four weeks to two years. Nine cases (75%) were diagnosed preoperatively as cases of infected sebaceous cysts. Resection and direct closure without flap were done for nine (75%) of them and in other three cases (25%), the PNS were resected and left for secondary healing. Three of the patients (25%) developed recurrence.

CONCLUSION: Intermammary PNS occurs in young, obese ladies with large breasts which are kept in tight brassieres. Resection and primary closure is the main method of management.

© 2017 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Pilonidal sinus (PNS) is a common, perianal, recurring, inflammatory condition caused by hair penetrating to the outer layer of skin (epidermis) [1]. It is a pus and hair containing cavity connecting to the skin surface by granulation lined tract [2]. The incidence of PNS is 0.07% and account of 15% of perianal diseases [3]. It usually occurs in age range of 10–40 years [4]. Female gender is affected less frequently than males by a ratio of one to three [4]. Hairiness, male sex, young age, prolonged sitting, deep navel and cleft and poor personal hygiene are among the predisposing factors [3]. It clinically presents as redness, pain and discharge [5]. Typically PNS occurs in the sacrococcygeal region. Rarely, it affects other areas of the body like groin, axilla, umbilicus, interdigital web, suprapubic area, nose, clitoris, prepuce, penis, or occiput [1,3,6–11].

Presentation, diagnosis and management of atypical PNS (PNS occurring in areas other than natal cleft) differ from sacrococcygeal PNS [1]. Ninety percent of all umbilical PNS reported in three neighboring countries of Middle East, namely Iran, Iraq and Turkey [12]. While classical presentation of a typical PNS is discharging sinus, all most all scalp PNS present as nodule [10]. Sacrococcygeal PNS is a clinical diagnosis which does not need simplest investigation while for diagnosis of atypical PNS, the correct diagnosis is the last one to be thought [1]. Conservative management is starting to grow regarding perianal PNS while for atypical PNS still operation is the standard therapeutic option [13].

Literature lacks information regarding intermammary PNS (iPNS). Up to date, only six cases have been reported [5,8,14–17]. The aim of this study is to report and discuss the presentation and management of twelve cases of pilonidal sinus occurring in intermammary area.

* Corresponding author at: Faculty of Medical Sciences, School of Medicine, Department Cardiothoracic and Vascular Surgery, University of Sulaimani, François Mitterrand Street, Sulaimani, Kurdistan Region, Iraq.

E-mail address: fahmi.hussein@univsul.edu.iq (F.H. Kakamad).

2. Method

2.1. Registration and ethics

The research registry number has been taken in accordance with the declaration of Helsinki – “Every research study involving human subjects must be registered in a publicly accessible database before recruitment of the first subject”. The approval for conducting of this study was taken from the Scientific and Ethical committee of university of Sulaimani.

2.2. Study design

This study is a retrospective multicenter case series. The cases were consecutive. The research work has been reported in line with the PROCESS criteria [18].

2.3. Setting

The institutions in which the cases were managed were academic, community and private practice setting. All of them located in Kurdistan. The patients were received and managed during last 12 years (from January 2005 to January 2017). The cases were followed up to date (first of July 2017, mean duration of follow up was 15 months). The data collection was done during two months (first of May 2017 to first July).

2.4. Participants

Inclusion criteria were all cases of iPNS admitted to either of six major tertiary hospitals. They were diagnosed clinically and/or pathologically. The socio-demographic and clinical data were taken from patients' medical records, health care providers and the patients themselves.

2.5. Pre-intervention considerations

All patients were prepared for general anesthesia. They were checked prior to operation for vital signs, hemoglobin level, and chest x.ray. They were examined and the area of proposed incision was marked (Fig. 1).

2.6. Types of intervention(s) deployed

All patients underwent operation under general anesthesia in supine position. Prior to intervention, they were nil by mouth for six hours and received intravenous antibiotics.

2.7. Peri-intervention considerations

During the operation, the patients were followed up by continuous electrocardiography (ECG) monitoring. Intravenous fluid in form of crystalloid was given according to vital signs. For patients who pre-operatively diagnosed as case of iPNS strategy of excision and laid open was used. For those patients who were not diagnosed as cases of iPNS excision and primary closure was performed (Fig. 2). For the latter, open drain was also put.

2.8. Who performed the procedure(s)

The procedure was performed by specialist with a team composed of senior house officers and nurses.



Fig. 1. Preoperative marking of the incision site, the area was marking according to the induration.



Fig. 2. Incision and primary closure without flap.

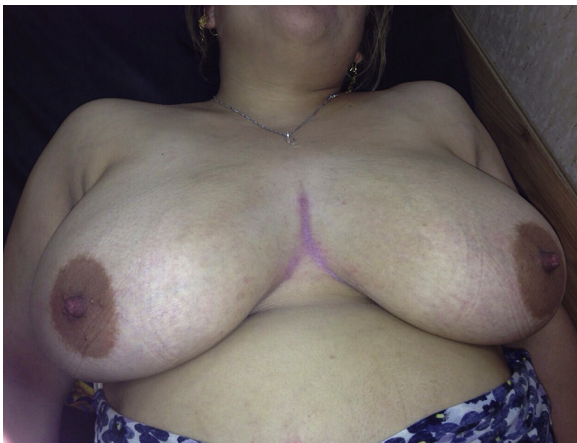


Fig. 3. Hyperpigmentation line in the intermammary region with large breast size.

2.9. Post-intervention considerations

Those patients who underwent only excision without primary closure were advised to do daily dressing until 21 days later on they should be seen the by specialists. The drain (if present) was removed after 14 days. The diagnosis of all cases was confirmed by the result of the histopathological examination of the excised specimen.

3. Results

3.1. Participants

The case series included 12 patients, all cases were female. The age range was from 13 to 29 years with mean age of 20.4 years. All of cases were obese with body mass index (BMI) ranged between 30.1–32.2 kg/m² with mean BMI of 30 kg/m². All of the patients had hyperpigmentation line between the two breasts as a marker of pressure on intermammary region (Fig. 3). All patients reported use of tight brassieres to decrease the size of the breast. The intermammary area was not found to be hairy in any cases. Three of the cases (25%) had history of perianal PNS. All of them presented with discharge for a long time ranging between four weeks to two years with mean of eight months. Definitive preoperative diagnosis was done for three (25%) of them. Others (75%) were diagnosed preoperatively as cases of infected sebaceous cysts. Resection and direct closure without flap were done for nine (75%) of them and other three (25%) were resected and left for secondary healing.

3.2. Outcomes and follow-up

Within 40 days of follow up, nine patients (75%) were found to be cured from the disease. Three of the patients (25%) developed recurrence. Two patients with recurrence were from those patients treated by excision and primary closure. These two patients were treated again by resection and primary closure. Third patient with recurrent iPNS was from those patients treated by excision and laid open. She was treated by re-excision and the wound was left for daily dressing and secondary healing.

3.3. Complications and adverse or unanticipated events

One patient developed bleeding 3 h after operation, controlled by compression and dressing. No any other significant complication was reported.

4. Discussion

PNS is relatively a common perianal problem while iPNS is an extremely rare disease with only six reported cases [3,5,8,14–17]. To best of our knowledge, this is the first case series. Previous studies showed that 67% of PNS occurs in male gender [2,8]. Regarding iPNS, there is no reported case in male gender [5,8,14–17]. All patients in this study were female. This may be explained by presence of breast in female gender. Hyperpigmentation in the region of intermammary area supports this claim. Typical and atypical PNS are disease of young patients with age around 15–30 years [1,13,14]. Regarding age of affection, iPNS is consistent with other type of PNS. Sunkara et al. like Salih and associates reported iPNS occurring in 16 years old female [8,14]. In this study, the age range was between 13–29 years with mean of 20.4 years. It was reported that hairiness is among the most important risk factor for developing PNS. However, this risk factor was not mentioned in case of iPNS in the literature [5,8,14–17]. The current study confirmed that being hairy is not necessary to develop iPNS as all cases had not have hairs at the area of the disease. Another well known risk factor for being affected by PNS is obesity [2]. This is consistent with the findings of this study (BMI of all patients were above 30 kg/m²). In this study, all cases reported to wear tight brassieres which was used for cosmetic purpose (to make breasts look smaller in size), although it is not addressed in previous case reports, theoretically tight brassieres increases the pressure posted by the breast on the intermammary region and enhance hair penetration into the skin. The most common presentation of PNS is chronic discharging sinus which was found in all cases of this study [13]. The diagnosis of perianal PNS is usually done by clinical examination, while atypical PNS is more problematic [1]. Apart from umbilical and interdigital PNS, all atypical PNS are least thought by physician during diagnostic work up for chronic discharging sinus [1]. Majority of our cases (75%) were not diagnosed as iPNS before operation, only 3 cases (25%) had been recognized as iPNS preoperatively. The most commonly used treatment modality for typical PNS is surgery which classically includes local excision and healing by secondary intention. However, post operative wound care following excision is difficult, leading to frequent and time-consuming work up [19]. Excision with primary closure is another alternative surgical therapy. The latter has the advantage of early cure rate, less job absence, more patient satisfaction and being less costly [20]. All surgical therapies put the patient at risk of morbidity and long time work absence, so they don't seem to be ideal management strategy [13]. Non-surgical therapies for PSD include phenol injection, laser ablation, and cream and glue application [13,21]. There are controversies regarding these treatment modalities as well. Qais et al. reported 55% recurrence after first trial of phenol injection [21]. Dag and associates reported 33% failure rate after three time phenol injection and 20% developed complication (13% burn, 3.5% cellulitis, 3.5% abscess) [22]. Management of atypical PNS is less problematic, as far as majority of them are not diagnosed preoperatively, they are managed by excision with primary repair [1]. Seventy five percent (nine patients) in this study were treated by excision with primary repair others (three patients, 25%) were treated by wide local excision and laid open for daily dressing and secondary healing. Not like sacrococcygeal PNS, iPNS has least technical problem with primary closure. The former needs flap for which countless techniques have been proposed in the literature [20]. However, flap has never been recommended for primary closure of iPNS. This may be explained by the fact that skin of the intermammary areas is much more lax and can be repositioned to cover the neighboring structures. The strength point of this study is that it is the first and unique case series reporting this number of patients with iPNS and in details highlight the presentation and management, while the limitation

is the rarity of the condition and small sample size which is difficult to reach decision making conclusion.

5. Conclusion

iPNS is another type of atypical PNS occurring in young, obese ladies with large breasts which are kept in tight brassieres. Resection and primary closure without flap is the main method of management.

Conflicts of interest

There is no conflict to be declared.

Funding

No source to be stated.

Ethical approval

Approval has been taken from Scientific and Ethical committee of university of Sulaimani.

Consent

Consent has been taken.

Author contribution

Tahir A Hawrami, Seerwan Hama Shareef and Abdulwahid M. Salih: surgeon performing the operation. Final approval of the manuscript.

Fahmi H. Kakamad, Hawbash M. Rahim, Dahat A. Hussein and Hunar A. Hassan: follow up of the patients, data collection and final approval of the manuscript.

Guarantor

Fahmi Hussein Kakamad.

References

- [1] A. Salih, F. Kakamad, R. Essa, M.S. Aziz, R.Q. Salih, S.H. Mohammed, et al., Pilonidal sinus of atypical areas: presentation and management, *PSJ* 3 (1) (2017) 8–14.

- [2] I. Sengul, D. Sengul, G. Mocan, Axillary pilonidal sinus: Fig. 1: histological appearance of pilonidal sinus (H&E stain), a case report, *N. Am. J. Med. Sci.* 1 (November (6)) (2009) 316–318.
- [3] A.M. Salih, F.H. Kakamad, Preauricular pilonidal sinus: the first reported case, *Int. J. Case Rep. Images (IJCRI)* 7 (3) (2016) 162–164.
- [4] K. Søndena, E. Andersen, I. Nesvik, J.A. Søreide, Patient characteristics and symptoms in chronic pilonidal sinus disease, *Int. J. Colorectal Dis.* 10 (1) (1995) 39–42.
- [5] P. Yavalkar, R. Shelke, Intermammary pilonidal sinus: a case report, *Indian J. Med. Case Rep.* 3 (2) (2014).
- [6] A.M. Salih, F.H. Kakamad, A case report of endoanal pilonidal sinus, *J. Case Rep. Images Surg.* 2 (2016) 60–62.
- [7] A.M. Salih, F.H. Kakamad, R.A. Essa, S. Othman, R.Q. Salih, M.S. Aziz, et al., Pilonidal sinus of the face: presentation and management – a literature review, *PSJ* 3 (1) (2017) 9–13.
- [8] A.M. Salih, F.H. Kakamad, M.H. Abdulqadr, Intermammary pilonidal sinus: a rare presentation, *Int. J. Case Rep. Images (IJCRI)* 7 (1) (2016) 48–50.
- [9] A.M. Salih, S.H. Mohammed, A.H. Dahat, Shvan H. Mohammed, Tariq H. Lhun, Postauricular pilonidal sinus; a case report with literature review, *Pilonidal Sinus J.* 3 (1) (2017) 4.
- [10] A.M. Salih, F.H. Kakamad, Scalp pilonidal sinus: a case report, *Int. J. Case Rep. Images (IJCRI)* 7 (3) (2016) 175–177.
- [11] A.M. Salih, F. Kakamad, I.J. Habibullah, M.H. Abdulqadr, Submental pilonidal sinus—the first reported case? *Pilonidal Sinus J.* 3 (1) (2017) 4.
- [12] Abdulwahid M. Salih, Fahmi H. Kakamad, Rawand A.Essa, Shvan H. Mohammed, Rawezh Q. Salih, Snur Othman, et al., Pilonidal sinus of the umbilicus; presentation and management. Article in press.
- [13] Hasan Mete Aksoy, Berna Aksoy, Didem Egeme Effectiveness of topical use of natural polyphenols for treatment of sacrococcygeal pilonidal sinus disease: a retrospective study including 192 patients, *Eur. J. Dermatol.* 20 (4) (2010) 476–481.
- [14] A. Sunkara, D. Wagh, S. Harode, Intermammary pilonidal sinus, *Int. J. Trichol.* 2 (July (2)) (2010) 116–118.
- [15] J. Lion-Cachet, Inter-mammary pilonidal sinus, *S. Afr. J. Surg.* 9 (July–September (3)) (1971) 141–142.
- [16] H.C. Richardson, Intermammary pilonidal sinus, *Br. J. Clin. Pract.* 48 (July–August (4)) (1994) 221–222.
- [17] E. Demiralay, A. Höbek, G. Altaca, Inter-mammary pilonidal sinus; an extremely rare location: case report, *BTDJMB* 5 (2) (2009) 78–79.
- [18] R.A. Agha, A.J. Fowler, S. Rammohan, I. Barai, D.P. Orgill, The PROCESS Group, The PROCESS statement: preferred reporting of case series in surgery, *Int. J. Surg.* 36 (Pt. A) (2016) 319–323.
- [19] A. Dwivedi, Management of pilonidal sinus by Kshar Sutra, a minimally invasive treatment, *Int. J. Ayurveda Res.* 1 (2) (2010) 122.
- [20] A.M. Salih, H.H. Ghalib, D.A. Ismael, M.I. Gubari, H.M. Hassan, M.S. Aziz, Outcome of rhomboid flap and Z-plasty in pilonidal sinus surgery, *JSMC* 5 (2) (2015) 1–8.
- [21] Q.K. Baqir, The role of phenol injection in the treatment of pilonidal sinus disease, *Bas. J. Surg.* 15 (March) (2009) 78–80.
- [22] (a) A. Dag, T. Colak, O. Turkmenoglu, A. Sozutek, R. Gundogdu, Phenol procedure for pilonidal sinus disease and risk factors for treatment failure, *Surgery* 151 (1) (2012) 113–117; (b) E. Elsey, J.N. Lund, Fibrin glue in the treatment for pilonidal sinus: high patient satisfaction and rapid return to normal activities, *Tech. Coloproctol.* 17 (1) (2013) 101–104.

Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.