

AN UNUSUAL PRESENTATION OF PILONIDAL SINUS – A DIAGNOSTIC CHALLENGE!

Othorhinoaryngology

Dr. Rakesh B.S*

Assistant Professor, Department of Othorhinoaryngology and head and neck surgery, JSS Medical college and Hospital, JSSAHER , Mysuru, Karnataka, India-570004.
*Corresponding Author

Dr. Kavya Sivapuram

Junior Resident, Department of Othorhinoaryngology and head and neck surgery, JSS Medical college and Hospital, JSSAHER , Mysuru , Karnataka, India- 570004.

Dr. Akash Grampurohit

Junior Resident, Department of Othorhinoaryngology and head and neck surgery, JSS Medical college and Hospital, JSSAHER , Mysuru , Karnataka, India-570004.

Dr. Sriram Madhan

Junior Resident, Department of Othorhinoaryngology and head and neck surgery, JSS Medical college and Hospital, JSSAHER , Mysuru , Karnataka, India-570004.

ABSTRACT

Introduction: Pilonidal sinus (PNS) is any subcutaneous sinus containing hair. 97.8% of PNS are in sacrococcygeal region making it the most common site of PNS. Extrasacrococcygeal PNS accounts for approximately 2.2% of PNS. Few cases of PNS of atypical sites have been reported. The purpose of this paper is to report an unusual case of pilonidal sinus over the submandibular region.

Case Report: A 36 year old male patient presented with a painless swelling over left side of neck since 2 weeks which on examination had no tenderness and no local raise of temperature. Ultrasonography of neck was suggestive of abscess. Excision of the swelling was done and histopathological examination showed chronic inflammation with foreign body giant cell reaction suggesting diagnosis of Pilonidal sinus.

Conclusion: Pilonidal sinus over submandibular region is unusual and a diagnostic challenge. Total excision and primary closure was done in this case.

KEYWORDS

pilonidal sinus, submandibular region, recurrent abscess, foreign body giant cell reaction

INTRODUCTION:

Pilonidal sinus (PNS) is any subcutaneous sinus containing hair¹. This was first described by Anderson in 1847². The term Pilonidal sinus (Latin, pilus—hair, nidus —nest) was coined by Hodges in 1880². It can be congenital and acquired. Penetration of hair fragments into the skin causes PNS³. Discharging non healing sinus and recurrent abscess are the commonest presentations of PNS¹. The classical site of PNS is sacrococcygeal area^{3,4,5}. Few cases of PNS of atypical sites like submental area, nose, umbilicus, scalp, perineum, face, finger web, axilla, intermammary region, clitoris, prepuce have been reported^{3,4,5}

Case Report:

A 36 year old male patient, auto driver by occupation, presented with complaints of a painless swelling over left side of neck since 2 weeks, there was no change in size of swelling and not associated with any symptoms. Patient does not have any comorbidities, history of similar complaints 2 years back for which incision and drainage was done.

On examination, a solitary swelling of size 3x2 cms, oval in shape noted over left submandibular region, skin over swelling is normal except for a scar. No discharging sinuses noted. On palpation, swelling is non tender, no local raise of temperature, cystic in consistency , mobile and skin over the swelling is not fixed.

Blood investigations turned out to be within normal limits, USG neck showed a well defined thick walled collection in deep subcutaneous plane of left submandibular region with multiple well defined linear hypoechoic structures within – suggestive of abscess.(Fig a)

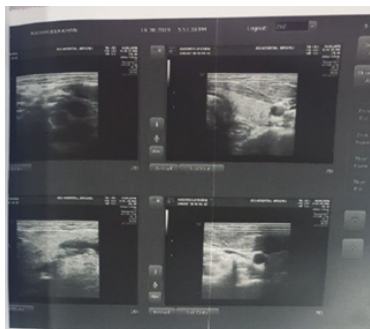


Fig a: Pre-operative USG neck showing features of abscess

Patient was planned for surgery, incision was placed over the swelling, Surprisingly a tuft of hair along with fluid noted. (Fig b,c) Swelling was removed in toto and a thorough curettage was done, and wound was closed in layers. Specimen was sent to histopathological examination. Meanwhile, patient was treated with higher antibiotics.

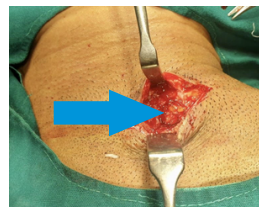


Fig b: Intra operative picture showing tuft of hair.



Fig c : Picture showing tuft of hair after removal.

Histopathological examination showed multiple fragments of fibro connective tissue infiltrated by chronic inflammatory cell infiltrate and foreign body type giant cells, features suggestive of chronic inflammation with foreign body giant cell reaction. (Fig d)

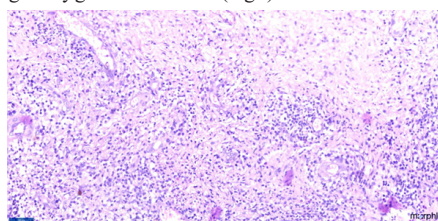


Fig d : Histopathological picture showing foreign body giant cell reaction

DISCUSSION:

Pilonidal sinus can be Sacrococcygeal and Extrasacroccygeal PNS. 97.8% of PNS are in sacrococcygeal region making it the most common site of PNS⁶ Extrasacroccygeal PNS accounts for approximately 2.2% of PNS and includes all the atypical sites of PNS⁶ like umbilicus, nose, chin, mandible, neck, scalp, face, post auricular area, hand, intermammary region, submental region⁷ Presence of loose hair in the cavity or sinus tract is the pathognomonic finding²

PNS in interdigital region was the first reported case of atypical PNS in 1942, followed by PNS of scalp in 1972 . A study by Salih AM *et al.* reported 9 cases of PNS on face, nose being the commonest site(4 cases), others include PNS of mandible, buccal area, pre auricular area, forehead¹

In another study done by Salih A *et al.*, a total of 302 cases of pilonidal sinus were reported in atypical sites⁵, and according to this study the most common atypical site was umbilicus (272 cases) which accounts for 90% of the cases in the study, and rest of the cases in hand(3.9%), scalp(1.7%), perianal region (1.3%), intermammary (1%), face(0.7%) , and reported cases of penis, clitoris, prepuce.(one each)³

Earlier literature supported that etiological origin of PNS is congenital. Of late, acquired theory of development of PNS is being well accepted². The acquired theory by Patey and Scarfe states that hair sucked into a sinus is infective in origin¹. Roger Brearley's acquired theory proposed that hair puncturing the intact skin leads to formation of sinus. Foreign body reaction caused by the hair forms a hair filled abscess cavity. PNS has male preponderance and is common after puberty which coincides with our case report^{8,9,10}

The commonest presentation of PNS is discharging sinus according to reports in literature by Salih AM *et al.*, 7 out of 9 cases of PNS of face presented with discharging sinus, one case presented with swelling and other case with abscess. Thus, PNS can also present with swelling or recurrent abscess. In our case report, patient presented with a non tender swelling over left submandibular region. O'Sullivan *et al.* reported a PNS in a 30 year old male who presented with swelling and tenderness at the left side of mandible for about 7 months duration⁷

The first case of PNS of neck was reported in 1992 by Miyata *et al*, 21 year old male patient presented with left nuchal abscess³. Meher *et al* reported second case of PNS, a 24 year old male patient with chronic discharging sinuses on the right side of upper neck for 3 years³. An another case report on PNS of neck was done in 2017 by Abdulwahid M. Salih *et al* , 20-year-old female presented with chronic multiple sinuses in the posterior part of neck³. This case report on PNS over left submandibular region will add up to the present literature on atypical sites of PNS.

On literature review, the standard method of management of atypical PNS was excision with primary closure under general anesthesia^{1,3,5} and a few case under local anaesthesia. In our patient also, excision with primary closure was done under general anesthesia, post-operative period was uneventful and no recurrence noted till date.

The histopathological picture of facial PNS does not differ from PNS of other areas including PNS of sacrococcygeal region. Presence of loose fragmented hair shafts with foreign body giant cells on histopathological examination gives us the diagnosis of PNS¹⁰

CONCLUSION:

PNS over submandibular region is rare and presents as a diagnostic challenge. Reports on atypical sites of PNS emphasize the need for considering atypical PNS as one of the differential diagnosis in patients presenting with discharging sinus and recurrent abscess. Total excision and primary closure will be the reasonable mode of treatment. The current case was managed with excision and primary closure.

Acknowledgement : We thank the institution, all the teaching and non-teaching staff who supported us for this article.

Conflicts Of Interest: No conflicts of interest among authors.

REFERENCES:

1. Salih, Abdulwahid & Kakamad, Fahmi & Essa, Rawand & Othman, Snur & Salih, Rawezh & Sleman, Masrur & Hussein, Shvan & Rahim, Hawbash & Hussein, Dahat & Hassan, Hunar. (2017). Pilonidal sinus of the face: presentation and management - a

literature review.. pilonidal sinus journal. 3. 9-13.

2. Paulose, K., Khalifa, S., Raj, S., & Saeed, T. (1989). Pilonidal sinus of the nose. The Journal Of Laryngology & Otology, 103(12), 1210-1213.
3. Salih AM, Kakamad FH, Salih RQ, Baba HO, Mohammed SH, Najar KA, Kakamad SH, Abdullah AN. Pilonidal sinus of neck: A case report. International Journal of Case Report and Images (IJCRI) 2018 ;9:100882Z01AS2018
4. O'Sullivan, M., & Kirwan, W. (2000). Post-traumatic pilonidal sinus of the face. British Journal Of Dermatology, 143(6), 1353-1353.
5. AM, Salih & Kakamad, Fahmi & RA, Essa & MS, Aziz & Salih, Rawezh & Hussein, Shvan & YA, Saeed & IJ, Habibullah & AS, Muhiudeen & S, Othman & HA, Mohameed. (2017). Pilonidal sinus of atypical areas: presentation and management. Pilonidal Sinus Journal. 3. 7.
6. Destek, S., Gul, V., & Ahioglu, S. (2017). Rare Type Cranial Postauricular Pilonidal Sinus: A Case Report and Brief Review of Literature. Case Reports In Surgery, 2017, 1-3.
7. Salih AM, Kakamad FH, Habibullah IJ, Abdulqadr MH. Submental pilonidal sinus: the first reported case. PSJ 2017; 3(1): 14-18.
8. Tomas, P. (2017). Pilonidal Sinus at Lateral Aspect of Neck. Archives In Cancer Research, 05(03).
9. Eryilmaz, R., Sahin, M., Okan, I., Alimoglu, O., & Somay, A. (2005). Umbilical Pilonidal Sinus Disease: Predisposing Factors and Treatment. World Journal Of Surgery, 29(9), 1158-1160.
10. M. Salih, A., & H. Kakamad, F. (2016). Preauricular pilonidal sinus: The first reported case. International Journal Of Case Reports And Images, 7(3), 162.