Bofares *et al.*
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CASE REPORT Open Access

Pilonidal sinus of external auditory canal as unpredicted lesion: rarest case presentation and review of literatures

Khaled Mohamed Bofares[1*], Huda M. Abdallah Hassan[2] and Mohammed Faraj Ali[3]

Abstract

Background Pilonidal sinus (PNS) is considered as a benign lesion of hair-bearing areas, particularly at the sacrococcygeal area. It is presented as a hole or tunnel in the skin. It may be filled with fluid or pus resulting in the formation of a cyst or abscess. Although PNS is a well-established lesion in general surgery as well as dermatology specialties by its occurrence at the sacrococcygeal region, but the surprising presentation of PNS is that at ENT specialty by its appearance at the external auditory canal (EAC), which is deemed unpredicted lesion at this particular region.

Case presentation Nine years female Libyan child has been presented with a history of persistent offensive bloody stained yellowish discolored left ear otorrhea a few months before her presentation. The local examination including the microscopic examination revealed evidences of a grayish-pinkish pedunculated polypoidal mass raised from the junction of the superior wall with the anterior wall of the EAC, at the site of the junction between the cartilaginous and bony portions of EAC. The temporal bone CT scan revealed evidences of radiolucent soft tissue density mass arising from the wall of the EAC at its anterior-superior junction, with the intact tympanic membrane as well as the middle ear cavity. This was most probably in favor of the suggestion of granulation tissue or aural polyp. The mass was completely excised as one piece from its pedicle and sent for histopathological evaluation, which was in favor of the diagnosis of PNS.

Conclusion We consider this case for reporting because of its scarcity and unexpected lesion at EAC. To the best of our knowledge, our case constitutes the second case of the EAC specifically, and the third case of external ear generally reported in the world. Thus, we tried to illustrate this unique and rare EAC lesion concerning how it is clinically as well as pathologically presented. In addition, we tried to review the literatures to elucidate whether this rare lesion has been reported before.

Keywords Pilonidal sinus, External auditory canal lesions, External ear lesions

Background

Pilonidal sinus (PNS) is a developmental benign lesion, which is commonly presented in hair-bearing areas exposed to continuous pressure friction. Thus, it is frequently seen at the sacrococcygeal region in between the buttocks. From the pathogenesis point of view, this lesion occurred through the interaction of three main predisposing factors: first is the presence of optimum levels of the sex hormones required for sufficient maturation of body sex criteria, which leads to the second factor that enhancement of the hair growth as a part of the normal

*Correspondence:
Khaled Mohamed Bofares
bofaresstat2012@yahoo.com
[1] Faculty of Medicine, Omar Almoukhtar University, AL-Beyda City, Libya
[2] Department of Pathology, Faculty of Medicine, Omar Almoukhtar University, AL-Beyda City, Libya
[3] SHO, AL-Beyda Medical center (AMC), AL-Beyda City, Libya

sex development. The third factor is a mechanical factor in form of the presence of frequent pressure friction at the zones of active hair growth. Therefore, the incidence of this lesion becomes higher at the puberty age group, among which there will be the highest activity of hair growth. In the presence of persistent pressure friction, the actively growing hair will be pushed and affected inward resulting in the formation of a hole or tunnel at the skin. This tunnel may be got filled with fluid or pus accumulation because of the lack of proper personal hygiene at these areas and superadded infection leading to abscess collection. The lumen of the sinus itself will provide a significant negative pressure that cause more hair nesting and furthermore tunnel dilation, which causes more drawing in more hair, therefore the cycle will be perpetuated [1–3].

Historically speaking, The term pilonidal was first given by R. M. Hodges in 1880, and it is a Latin term, pilo for hair and nidal for nest, thus pilonidal a phrase that described the gross pathological morphology of this lesion as a nest of hair. For the first time, this lesion was discovered during Second World War among young soldiers who were exposed to the long-standing pressure frictions on their bottoms during prolonged war Jeep cars deriving, therefore at that time, this lesion was named as Jeep rider's disease or Jeep bottom disease [4, 5].

In addition, from the epidemiological point of view, the incidence rate of this condition is 26:100,000. The males are more affected than the females; the male-to-female ratio is 3-4:1. In accordance, the young men were showed the highest incidence for this lesion, and the age of highest incidence was 20 years [5–8].

The PNS constitutes an important clinical entity; it may be associated with recurrent attacks of infection, because it usually occurs at the dirtiest high-humidity zones of the body, and with a lack of proper local personal hygiene and care, this will lead to the persistence of purulent discharging sinus. In addition, as a rare situation of PNS, the constant chronicity of the infection may lead to the change of the benign keratinizing squamous cells to malignant cells resulting in the development of squamous cell carcinoma [8–12].

The management of PNS is surgical. Patients with mild to moderate disease can be treated by simple techniques like the Bascom-1 procedure, which can be done under sedation and local anesthesia. Other procedures include Munro Z-plasty, Karydakis procedure and its Kitchen and Bascom-2 modifications, and En bloc excision of the cyst. In addition, the Limberg flap technique was very useful too [13–17].

Although the sacrococcygeal region was the commonest site for the development of this lesion, but also this lesion was diagnosed at the scalp, neck, axilla, around the umbilicus, pubic region, groin, penis, and interdigital web region. However, the external ear is deemed the rarest site for the occurrence of this lesion, up to our knowledge, there were two cases of pilonidal sinus of the external ear recorded in the world, one at the ear lobule and the other one at the EAC. Thus, our case can be considered as the second case of pilonidal sinus that was diagnosed in the world [1, 6].

Indeed, this case presentation becomes unique because it is not in line with the usual and well-established presentation of this lesion. Thus, we tried via this case report to illustrate invaluable aspects of this case, which makes the lesion unforeseen.

Case presentation

Nine years female Libyan child has been presented to ENT Outpatient Department, AMC, AL-Beyda City, Libya, with a history of persistent bloody stained yellowish discolored left ear otorrhea a few months before her presentation. The ear discharge was more watery rather than muco-purulent; it has no relation to upper respiratory tract infection. The discharge has an offensive odor. It was proportionally decreased by the administration of systemic as well as local antibiotics, which were prescribed by general practitioners. In addition, the discharge during its active perfusion was associated with otalgia that may increase in its severity to disturb the sleeping of the child. The social history revealed the poor socio-economic status of the child's family and the family was living in poor sanitation. There was no other significant history neither medical nor surgical.

On the examination, in the general examination, there were no significant findings.

The local examination including the microscopic examination revealed evidences of a grayish-pinkish pedunculated polypoidal mass raised from the junction of the superior wall with the anterior wall of the EAC, at the site of the junction between the cartilaginous and bony portions of EAC. The mass was soft in its consistency and bleed on touch. No other significant finding rather than the blood-stained watery purulent discharge with an offensive odor. The tympanic member was obscured by the mass and cannot be fully assessed.

The patient was investigated by a temporal bone CT scan, which revealed evidences of radiolucent soft tissue density mass arising from the wall of the EAC at its anterior-superior junction, with the intact tympanic membrane as well as middle ear cavity. This was most probably in favor of the suggestion of granulation tissue or aural polyp (Fig. 1).

The decision of the excisional biopsy of the mass was taken, which was done under local anesthesia. The mass was completely excised as one piece from its pedicle

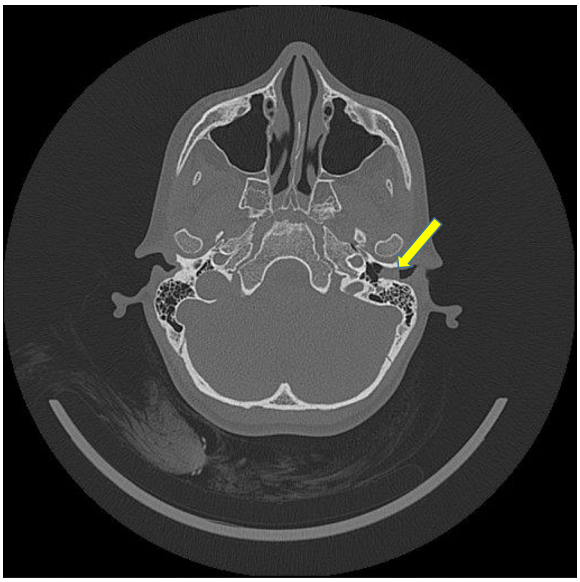

Fig. 1 Axial CT scan view of the skull corresponding to the temporal bone region showing the lesion at EAC (yellow arrow) with obvious intact tympanic membrane, middle ear cavity, and well-pneumatized mastoid air cells

and sent for histopathological evaluation. The tympanic membrane was assessed after the excision of the mass and it was completely normal.

The histopathological evaluation revealed a light micrograph of a polypoid structure covered by squamous epithelium with reactive epithelial changes and containing granulation tissue formation along with granulomatous inflammation surrounding the hair shafts. In addition to the demonstration of foreign body, type giant cells, and chronic inflammatory cells infiltrating the surrounding hair shafts, this histopathological picture was in favor of the diagnosis of PNS (Figs. 2, 3, and 4).

The patient received local tobramycin antibiotics with dexamethasone eardrops for three weeks and oral analgesics on need as postoperative medication. The postoperative follow-up sessions were 2, 4, and 8 weeks after the procedure, which showed complete normal healing of the skin at the site of the lesion without any stenotic lesions or granulation.

Discussion

PNS is a developmental lesion with well-established etiological as well as pathological criteria. Uniquely, our reported case did not follow these published concepts regarding PNS. We tried to discuss all the variant aspects that make our presented case phenomenal from the usual presentation of PNS.

First, from the epidemiological point of view, as it was mentioned in the introduction that PNS is of higher

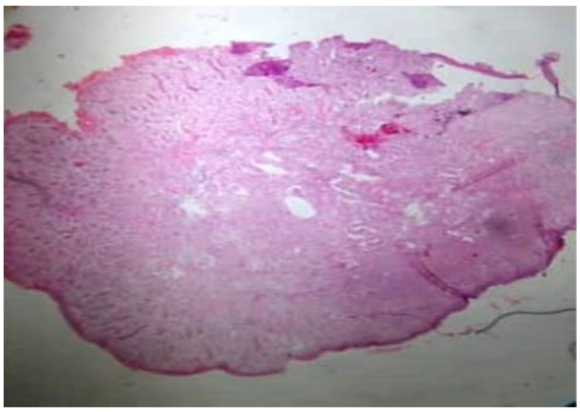

Fig. 2 Light micrograph of the polypoid structure covered by squamous epithelium with reactive epithelial changes and containing granulation tissue formation along with granulomatous inflammation surrounding the hair shafts (H&E stain; ×4)

incidence among adolescent males. In our case report, the patient was a girl in the childhood age group. This epidemiological presentation is completely varying from the well-known epidemiological criteria of PNS.

Moreover, there are common sites where this lesion was registered. In our case, the presentation was at the rarest site, which is EAC.

By the interpretation of our case criteria with the accredited characters of PNS as general, we can reach the fact that the pathogenesis of PNS is not depending upon the excessiveness of the hair growth activity as a part of secondary sex characters maturation at puberty age due to high levels of sex hormones excretion. In the same context, the long-standing local pressure frictions are not a significant etiological factor for the development of

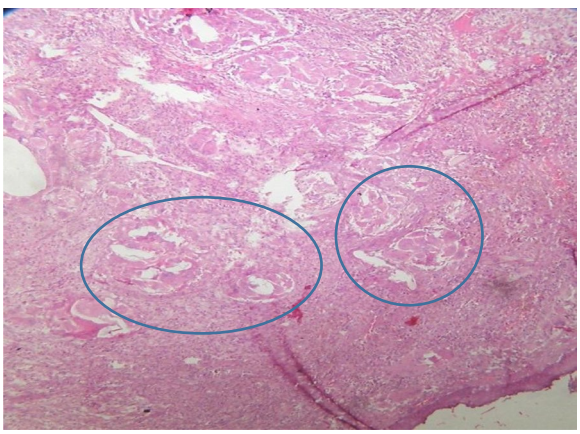

Fig. 3 Granulomatous inflammation (blue circles) composed mainly of foreign body type giant cells and chronic inflammatory cells infiltrate that surrounding hair shafts (H&E stain; ×10)

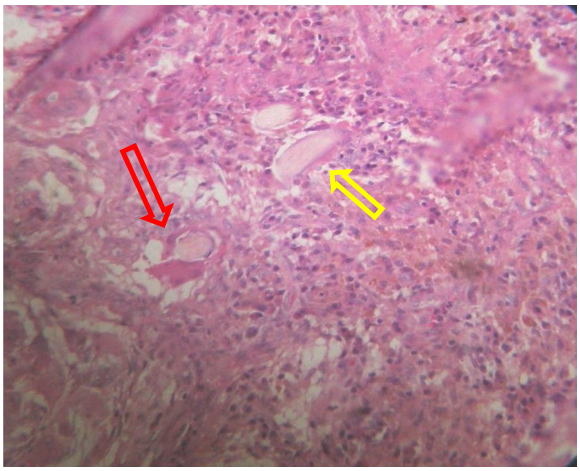

Fig. 4 Granulomatous inflammation composed mainly of foreign body type giant cells (red arrow) and chronic inflammatory cells infiltrate that surrounding hair shafts (yellow arrow) (H&E stain; ×40)

PNS. This can be proved by our reported case who was before puberty, and the lesion was manifested at a still yet non-haired region with no possible pressure friction effect.

The only acceptable explanation for the pathogenesis of this lesion among this child via the possibility of the foreign hair impaction at the EAC as the result of the scalp hair loss either spontaneously or during hair combing or trimming, subsequently as the immune response to this foreign body effect, the inflammatory granulomatous lesion will occur. This could be the same explanation for the pathogenesis of PNS among the barbers as well as animal hair trimmers, who developed this lesion at interdigital regions as the result of the pushing and impaction of the trimmed hair inward as foreign bodies. This will enhance and induce the reactive inflammatory granulomatous lesion, and it will be manifested as PNS [2, 3, 7].

The development of the squamous cell carcinoma on top of the PNS is very rare; this could be explained by the interference to excise this lesion before the dysplastic changes of the cells take place. The cellular dysplastic changes of the PNS tract cells are mainly time-dependent changes, i.e., by the prolongation of the duration of the chronicity of the infection; this will increase the risk of these malignant changes. Usually, the patients are managed by the excision of the lesion due to the persistence of the discharge before the dysplasia occurrence [8–10].

Therefore, our case can be considered as a singular case in the world as compared to the other case, which was reported in 2015 because of:

1- Our patient was female in sex as compared to the other case, who was a boy [1].

2- Among our patient, the lesion was at the left ear, while the other reported case presented with a right ear lesion [1].

Conclusion

Although the PNS is considered as the rarest inflammatory lesion of EAC, but should be suggested at the bottom of the differential diagnosis of the granulomatous lesion of EAC, which not responding to the ordinary protocols of management, particularly among childhood as well as adolescent ages. Indeed, further investigations are required to postulate why this lesion at EAC is confined to children rather than adult ages.

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Authors' contributions
KMB was the major contributor in writing the manuscript, in addition he was the surgeon, who operated the case and performed postoperative follow up sessions for her. HMAH was the pathologist, who performed the histopathological evaluation for the excised specimen. MFA was the SHO, who received the case at OPD and prepared her preoperatively. All authors read and approved the final manuscript.

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Availability of data and materials
The patient's data are documented and available in the patient's file.

Declarations

Ethics approval and consent to participate
The endorsement and statistical department at AMC, AL-Beyda City, Libya, ethically approved the publication of this case.

Consent for publication
Written informed consent for publication of the patient's clinical details and clinical images was obtained from the patient's father.

Competing interests
The authors declare that they have no competing interests.

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