Submental pilonidal sinus - the first reported case?

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ORIGINAL ARTICLE
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ABSTRACT
Introduction: Pilonidal sinus (PNS) usually affects the sacrococcygeal region. To our knowledge, there is no reported case of submental PNS. The aim of this paper is to report possibly the first case of submental PNS.

Methods: Case report.

Results: A 22 year old female presented with a submental mass for 7 years which proved to be a diagnostic challenge. From the sonographic point of view, it was diagnosed as a dermoid cyst, clinically diagnosed as a thyroglossal cyst while histopathological examination suggested the diagnosis of PNS.

Conclusion: Submental PNS could occur and present with diagnostic challenge. Complete resection with direct closure was performed.

Keywords: face, submental, pilonidal sinus, dermoid cyst, thyroglossal cyst

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INTRODUCTION
Pilonidal sinus (PNS) is a subcutaneous sinus which contains spontaneously induced by hair (1). Although previously referred to as a congenital condition, it is now regarded as acquired condition. It may present as a (recurrent) abscess or as a non-healing, discharging sinus (2). PNS usually affects the sacrococcygeal region. However, it may rarely occur in other regions like groin, axilla, umbilicus, interdigital web, clitoris, suprapubic area, scalp, prepuce, endoanal canal, intermammary region, occiput, penis (3-9). PNS involving the face is a very rare variant of the disease (10). To best of our knowledge, there is no reported case of PNS involving the submental area. We reported a young female who may have presented with submental PNS.

METHODS
Case report.
RESULTS
A 22 year old female presented with painful submental mass for 7 years. The patient had history of an operation at the same area 6 years previously. On local examination, a tender mass was found at submental area, measuring about 2 x 2 centimeters, attached to the skin without an external opening. Blood tests were within normal range. Ultrasound of the neck showed a well defined, hyperechoic lesion measuring about 20 x 18 x 15 mm with multiple hypoechoic foci - suggestive of a dermoid cyst.

Under general anesthesia an elliptical incision was performed and a tract like thickening was felt inside the lesion, making the picture more likely to be thyroglossal cyst. Complete excision of the mass with partial removal of hyoid bone was performed. The wound was closed directly using polypropylene 2/0.

On histopathological examination a tract infiltrated and surrounded by multiple, mixed inflammatory cells was identified. This was associated with a foreign body giant cell reaction and hair fragments (Figure 1) suggesting pilonidal sinus.

Because of this unusual histological diagnosis (especially when surgeon suspected a thyroglossal cyst) examination by a second pathologist was requested, again pilonidal sinus was diagnosed. The postoperative course was uneventful. Two years later, there was a white scar with clear margin.

Figure 1: Histopathological examination of the specimen showing foreign body giant cells (black arrow) with hair fragment (red arrow)

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DISCUSSION

PNS is a complex disease that causes both embarrassment and discomfort to the patient. It has a direct negative cost effect to the healthcare system and indirect negative cost effects to society through work absence (9). Facial PNS is very rare. Salih et al.’s literature review identified nine reported cases of facial PNS (10). Females with facial PNS are even rarer, among the nine cases reviewed, only one was female - 24 year old with forehead PNS reported by Tanwar et al at 2014 (11). Among facial PNS, nose PNS is the most common location while submental PNS has never been reported - that is why the team did not consider PNS in the current case (10).

The etiology of PNS is a matter of debate. Regarding sacrococcygeal PNS, congenital origin was favored initially but this was rejected by authors in the second half of the twentieth century (9). Friction (adduction - abduction), suction, shaving massage, pounding, maceration and minor infection are proposed mechanisms playing a role in the acquired theory of PNS (12).

The histopathological features PNS have been discussed in detail previously. Almost all authors accept that the presence of loose hairs is necessary for the diagnosis of PNS. Diagnosis of PNS in sacrococcygeal area is straight forward most of the time. PNS at atypical areas may present with diagnostic challenges (1).

The medical records for this patient’s first operation could not be retrieved but she stated the first presentation was the same as the second. The radiologist put a dermoid cyst as the first differential diagnosis based on the sonographic features. The surgical team considered thyroglossal cyst as the diagnosis. The argument for the latter was the site, being recurrent, and most importantly presence of a palpable tract intra-operatively. We performed complete excision of the mass and partial resection of the hyoid bone and placed a drain which remained in place for seven days. Two pathologists reviewed the histology. Excision and direct closure of the wound (with or without drainage) is accepted management of facial PNS (10).

In the literature review of facial PNS, there were no reported cases of recurrent facial PNS (10). This case is controversial as it is at an unusual site and recurrent. Lots of unequivocal hairs are not present in the histological section. Pathologists have not mentioned whether thyroid tissue was present in the adjacent connective tissue. With a previous operation at this site, the material demonstrated may have been implanted from this operation.

In summary, submental PNS is exceedingly rare and presents with a diagnostic challenge. Complete resection with direct closure seems a reasonable mode of therapy.

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CONTRIBUTIONS
Abdulwahid M. Salih: substantial contribution to the concept and design, drafting the manuscript, final revision and approval of the manuscript, the surgeon performing the operation.

F.H. Kakamad and Imad J. Habibullah: substantial contribution to the concept and design, drafting the manuscript, final revision and approval of the manuscript, follow up the patient and data collection.

Mustafa H. Abdulqadr: substantial contribution to the concept and design, drafting the manuscript, final revision and approval of the manuscript and histopathological examination of the specimen

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