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CASE REPORTS Open Access

Atypical compound melanocytic naevus of the urethral meatus case report

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Abstract

Background Atypical melanocytic naevus (AMN) of the urethral meatus is very rare with only a handful of reported cases. It is important to note that the rarity of AMN in the penis may contribute to a delay in diagnosis and treatment, as well as a lack of awareness among healthcare providers. This is the first case report to describe workup and management of AMN of the urethral meatus of a male, making it a valuable contribution to the literature.

Case presentation A Caucasian male patient in his fourth decade presented with a several month history of a pigmented lesion on his glans penis. The differential diagnosis was of malignant melanoma but after biopsy and specialist pathological review, atypical melanocytic naevus was diagnosed. A punch biopsy, followed by two wide local excisions, was undertaken for complete excision and evaluation of the lesion.

Conclusion Since AMN cannot be clinically distinguished from malignant melanoma, timely histological evaluation of the lesion is essential for workup. In this case, site and visibility of the lesion, expedient biopsy, and tissue analysis resulted in identification and excision of the naevus early enough for penis preserving surgery to be performed.

Keywords Urological surgery, Dermatology, Case report

1 Background

Atypical melanocytic naevi are acquired pigmented melanocytic proliferations. They most commonly occur in sun-exposed areas, such as the trunk, scalp, and forehead [1]. It is very rare for them to appear on the glans penis and even rarer to present at urethral meatus, with only a small number of cases reported [2]. We present a case of atypical compound melanocytic naevus of the urethral meatus, which raised concerns about melanoma and posed difficulties in clinical management.

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2 Case presentation

A man in his fourth decade of life presented to the dermatology clinic with a pigmented lesion on his glans penis (Fig. 1). He had noticed the lesion a few months earlier, and no associated symptoms were reported. He had no other medical history of note, was fit and well, and smoked 10 cigarettes a day. On examination, a 4-mm pigmented lesion was observed at the junction between the glans skin and the urethral mucosa. It had spread around the urethral meatus and extended 2 mm into the urethra. The patient had no lymphadenopathy or hepatosplenomegaly.

An urgent punch biopsy revealed severely atypical melanocytic proliferation, which was reported to be highly concerning for vertical growth phase malignant melanoma with a Breslow thickness of at least 1 mm. A computed tomography (CT) scan of the chest, abdomen, and pelvis demonstrated no convincing evidence of nodal or metastatic disease. The punch biopsy was re-reviewed at a regional penile cancer multidisciplinary team meeting (MDT), which reported a compound melanocytic lesion

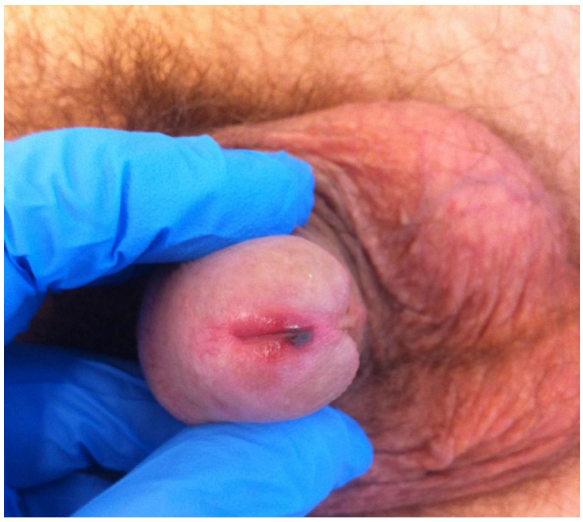

Fig. 1 Atypical melanocytic naevus of the urethral meatus. Photograph of a pigmented lesion on the glans penis of a male subject. The lesion was biopsied to determine the cause of the discoloration. This photograph documents the location and appearance of the lesion prior to biopsy, providing valuable visual information for clinical assessment and diagnosis

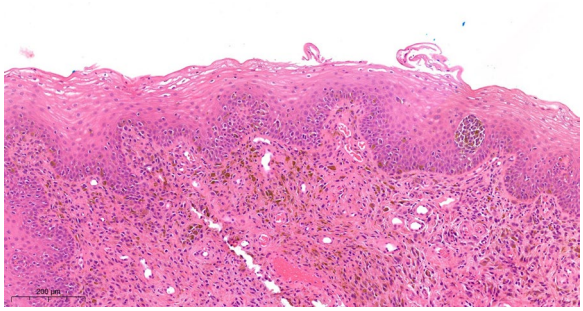

Fig. 2 Atypical melanocytic naevus. Cross-sectional histology slide of a pigmented penile lesion in a male subject, stained with hematoxylin and eosin (H + E). The slide shows the characteristic layers of the skin, including the epidermis and dermis. The junctional (junction between epidermis and dermis) and dermal components contain pigmented (brown) cells with large nucleoli and atypical outlines consistent with atypical melanocytes with severe dysplasia

with severe dysplasia in keeping with an atypical melanocytic naevus showing melanocytic proliferation and atypia in a lentiginous pattern associated with lymphocytic infiltrate (Fig. 2). Complete excision was advised; onward management was under the penile cancer team.

Following this MDT, upon clinical review, there was a very small pigmented lesion visible at the site of the previous biopsy at the meatal edge. Wide local excision was recommended. This had positive margins for dysplastic cells; therefore, additional wider local excision was performed. The patient was kept under close clinical review, and at 1 year after the initial operation, there was no sign of recurrence, in keeping with a benign lesion. The patient had no urinary symptoms before or after treatment.

3 Discussion

Atypical melanocytic naevi are skin lesions whose clinical and histological features lie somewhere on the proliferative continuum from a common mole to a melanoma [3].

They are common in Caucasian populations, with a prevalence between 2 and 5% [3]. They are rare in black, Asian, or Middle Eastern populations. AMN typically occurs in the sun-exposed areas of the torso, face, arms, and legs. It is rare on the penis—the incidence of AMN of the urethra is unclear; only six cases involving the meatus have been reported in the literature [4].

An AMN can develop throughout an individual's lifetime. Those with sporadic atypical naevi typically have a relatively small number of lesions, whereas those with a genetic cause may have a large number of moles. The molecular events that direct melanocytic proliferation in the pathogenesis of AMN formation are unknown, but they appear to include a constellation of genetic, epigenetic, and environmental factors (e.g., B-RAF mutation status, cell cycle protein expression, histone modifications, and ultraviolet radiation exposure), which may regulate cellular mechanisms that bypass cellular senescence [1]. Mutations in CDKN2A (p16INK4a) can result in uncommon autosomal dominant familial atypical multiple mole syndrome [1]. AMN should be considered as intermediate lesions of tumor progression, as approximately 30% of melanomas arise in association with AMNs. The risk of transitioning to melanoma depends on the degree of atypicality, ranging from mild to severe [1].

On examination, AMN presents with irregular, ill-defined borders, variable tan to dark brown pigmentation, and an erythematous base. The clinical differential diagnoses of penile AMN include common naevi, malignant melanoma, Spitz naevi, melanotic macules, angiokeratomas, seborrheic keratosis, squamous cell carcinoma, and basal cell carcinoma. Most cases of penile AMN present with a lesion; however, the diagnosis of AMN or other pigmented penile lesions, like melanoma, can be delayed if the lesion is out of sight (e.g., urethra) or if there is some reluctance to consult a physician due to the sensitivity of the area. As a result, these cases can present with symptoms of more advanced disease such as dysuria, obstruction, hematuria, and discharge [5].

Penile AMN is clinically indistinguishable from malignant melanoma. Therefore, in order to establish a definitive diagnosis and to avoid extensive surgery (which may consist of glansectomy or partial or radical penectomy),

an excisional biopsy with subsequent histological examination should be performed [4].

More limited approaches (punch, shave scoop biopsies) will likely require subsequent complete excision based on the advice of the pathologists (as in this case), so residual parts of the lesion can be reviewed [6]. Therefore, initial complete or wide excisions of atypical naevi reduce the likelihood of surgical reinterventions. There are other diagnostic techniques available such as reflectance confocal microscopy, which may be a valuable noninvasive alternative to a partial biopsy (e.g., punch) for 'initial diagnosis' where complete excision as a first intervention is not feasible.

AMN can be managed less radically than malignant lesions, with far less impact on the penile function. This is particularly relevant for urethral meatus lesions, as a more conservative approach will have less impact on urinary function and urethral scarring.

Melanoma of the penis and urethra is rare, with less than 200 cases of primary urethral melanoma reported since first described [7]. The peak incidence of penile melanoma is in the sixth to eighth decades of life, and the prognosis is generally poor. Clinically, melanoma presents as an approximately 1 cm blue-black to reddish-brown pigmented papule plaque or ulceration. Penile melanoma may be localized on the glans (55%), prepuce (28%), penis shaft (9%), or urethral meatus (8%) [2]. Expedient follow-up of patients with clinical and dermoscopic features of urethral AMN is essential for the early identification of any lesion transformation (to melanoma) and timely surgical management.

4 Conclusion

Atypical melanocytic naevus of the urethral meatus is a rare presentation that requires multidisciplinary team management. AMNs are clinically indistinguishable from malignant melanoma; therefore, after thorough history and examination, the lesion should be biopsied for histological assessment. Although it may be tempting to perform a limited biopsy for cosmetic and sensitivity reasons, complete, or wide local excision is necessary for absolute evaluation of the lesion. Differentiating AMN from melanoma means less extensive surgery, preserving penile function and anatomy, improving the outcome for the patient. While AMNs can arise at any age, malignant melanoma of the penis is more common in the sixth to eighth decades of life.

Abbreviations

AMN Atypical melanocytic naevus
CT Computed tomography
MDT Multidisciplinary team meeting

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J.M-W. sourced the clinical information and wrote the manuscript. SB contributed to and edited the manuscript and sourced and edited the clinical and histological images. RR and SD undertook critical revision for important intellectual content. All authors reviewed and approved the final version of the manuscript for submission.

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Competing interests
The authors declare they have no competing interests.

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