Verrucous carcinoma arising in the setting of postnephrectomy sinus

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ABSTRACT

Marjolin's ulcers are malignancies that arise in chronic venous ulcers, scars, burns, long-standing wounds or sinuses. The most common type of carcinoma arising in the Marjolin's ulcer is squamous cell carcinoma (SCC), followed by basal cell carcinoma. Verrucous SCC (VSCC) comprises about 3% of all SCCs. It is most commonly diagnosed in the oral cavity, and the incidence is approximately one in a million/year. To the best of our knowledge, malignancy arising in a postnephrectomy sinus has never been described before. We herein report a case of VSCC in a sinus developed after nephrectomy done for renal stones.

Key words: Postnephrectomy, sinus, verrucous squamous cell carcinoma

INTRODUCTION

Malignant transformation in chronic ulcers is a well-recognized phenomenon.^[1] However, malignancy arising in the setting of long-standing fistula is a rare phenomenon. One such case arising from long standing colocutaneous fistula has been described in the literature.^[2]

Verrucous squamous cell carcinoma (VSCC) is a highly differentiated type of squamous cell carcinoma (SCC). Also known as Ackerman's tumor, it is composed of an exophytic, warty tumor with multiple filiform projections, which are thickened and club-shaped, and lined by well-differentiated stratified squamous epithelium. Most of the verrucous carcinomas have been described in oral cavity and larynx is followed by sinonasal tract and nasopharynx. [3] Although no specific anatomic location is exempt from this neoplasm, but verrucous variant of SCC arising in a long-standing fistula has never been described before.



CASE REPORT

A 49-year-old male presented with nonhealing wounds over the right flank for the past 2 years. The patient had undergone right nephrectomy for nonfunctioning kidney secondary to renal stones 2 years back. Histopathological examination of nephrectomy specimen revealed chronic pyelonephritis without any evidence of malignancy. General and systemic examination revealed no significant abnormality except for pallor. Hemoglobin was 6.6 g%. Rest of the hematological and biochemical investigations including renal function tests were within normal limits. He was nonreactive for HIV, HBs antigen and anti-HCV antibody.

Magnetic resonance imaging revealed an irregular hyperdense collection on T2 fat suppressed L3 images in right paraspinal muscles and retroperitoneum on L2-L3 level with a tract descending from it posteriorly and inferiorly up to subcutaneous plane [Figure 1]. Exploratory laprotomy was done, which revealed multiple sinuses reaching up to the anterior aspect of paraspinal muscle with collection of cheesy content in the tract. Sinus tracts were traced out, and cavities were curetted out after evacuation of collection.

We received excision biopsy of the sinus tract with attached soft tissues measuring together $7 \times 3.5 \times 2$ cm [Figure 2a]. Sinus tract was seen communicating with the skin on further dissection. Histopathological examination revealed hyperplastic stratified squamous epithelium

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growing as club-shaped/papillary formations with a pushing or bulldozing margins and an intact basement membrane [Figure 2b]. The epithelium was well differentiated without any evidence of nuclear pleomorphism or mitotic activity [Figure 2c]. Any foci of invasion or atypia could not be demonstrated despite extensive sampling. A diagnosis of verrucous carcinoma was rendered.

The postoperative period was uneventful, and the patient is still under follow-up

DISCUSSION

Squamous cell carcinoma can develop in the setting of chronic ulcers, scars, wounds, sinuses, and fistulas. The latent period is long (around 37 years) in patients with burn scars except for immunocompromised patients where it can develop as early as 1–7 years. Malignant tumors arising from previously existing fistulas are rare. [2] The incidence of carcinoma arising in pilonidal disease, anal fissure and osteomyelitic fistula is < 0.1%, 0.1%, and 0.2% respectively. [4] The development of malignancy in unhealed wounds can be attributed to chronic irritation and infection. [2] Regular biopsy of sinus tracts is necessary. A high index of suspicion will facilitate the early detection of malignant transformation. [1]

Verrucous carcinoma was first recognized in 1941 by Friedell and Rosenthal. They reported VSCC in eight patients with oral cavity tumors. It is a highly differentiated variant of SCC, is slow-growing and locally invasive but lacks metastatic potential. However, some of them might be locally aggressive. [5] The most frequent site is the glottis region of the larynx in the head and neck areas, but may develop in another mucosal surface of the ear, nose or throat. The characteristic histologic features are highly keratinizing surface, papillary areas and apparent absence of dysplastic change in squamous epithelium. [6]

Regional lymph node metastasis and distant metastasis are extremely rare. Surgery in resectable lesions offers the best prospect for long-term survival. ^[6] However, in patients who are not good surgical candidates, radiation therapy should be considered. ^[3]

Lee *et al.* reported a case of SCC in an iatrogenic colocutaneous fistula which developed 30 years after surgical removal of left renal stones.^[2] The SCC arising in the setting of renal stones has also been reported.^[7] However, there is no prior reported case of VSCC arising in long-standing sinus or fistula. Moreover, postnephrectomy sinus has never been described as a site of Marjolin's ulcer.

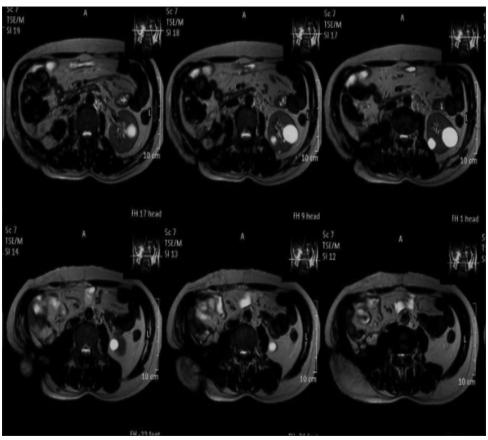


Figure 1: T2 fat suppressed magnetic resonance imaging revealing an irregular hyperdense collection in right paraspinal muscles, retroperitoneum on L2-L3 level with a tract descending from it posteriorly and inferiorly up to subcutaneous plane

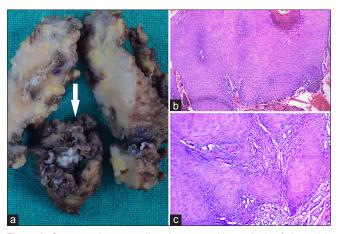


Figure 2: Gross specimen revealing sinus tract with attached soft tissue (a). Histopathological examination revealed hyperplastic stratified squamous epithelium growing as club-shaped fingers with a pushing margin and an intact basement membrane (b). The epithelium was well differentiated without any evidence of nuclear pleomorphism or mitotic activity (c)

Our patient presented with an unusual variant of SCC developing at a rare site.

CONCLUSION

We hereby report this case in order to alert the surgeons and pathologists regarding the existence of this rare, although detrimental development in long-standing fistulas. It is important to realize that any fistula/sinus should not be left unattended. A high index of suspicion should always be exercised in such cases and an urgent and aggressive therapeutic strategy should be instituted as soon as possible in the rare, but potentially malignant cases to save the patient from further catastrophic consequences.

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