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Letter to the Editor

Squamous Melanocytic Tumour at an Unusual Site: An Uncommon Case and Literature Review

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ABSTRACT

We herein present a case report of a 60-year-old male patient diagnosed with squamous-melanocytic tumour (SMT) in his anal region, comprised of two intermingled different phenotypic lesions. This dual tumour is quite uncommon and could potentially be challenging on both diagnose and also management. It is the first case reported on this location, moreover, the lack of consensus for these lesions makes it difficult to classify them. We reviewed the literature of similar SMT's and discussed its histogenesis. As they are rare, their biological behavior and potential metastasis remain unclear. Therefore, close follow-up is advised.

Keywords
Combined neoplasia; Squamous-melanocytic tumour; Uncertain; Anal canal.

INTRODUCTION

Anal squamous cell carcinoma is a rare condition. Of all cancers of the lower gastrointestinal tract, anal squamous cell carcinoma accounts for 4% of the total.[1] Likewise, primary malignant melanoma of the anorectal area are quite infrequent and only represents 0.4%-1.6% of all melanomas and less than 1% of anal canal tumours.[2] Not surprisingly, a combination of both (squamous cell and melanocytic lesions) is an exceeding rare tumour. Our case is the first SMT case reported in the anorectal region. The literature review shows nineteen SMT reported cases. Most of them located on the head and neck[3] and none in the perineal or genital areas. The biological behaviour remains unclear and albeit eighteen cases have not shown any further malignant report there is one case with SMT metastasis5. There is no definitive risk factor for SMT lesions although some of them have been linked to previous burn and solar conditions.[3-5]

CASE REPORT

A 60-year-old male patient presented a dark pigmented nodule on his anal canal. No other clinical information was provided. The clinical hypothesis was either a malignant or benign pigmented lesion. The lesion was excised through a shave biopsy and sent to histopathological analysis.

Histopathological Findings

Histological examination showed on low-power magnification a fairly well-circumscribed epidermal tumour with endophytic proliferation displaying striking dark pigment within it. The neoplasia surrounded adnexal structures such as hair follicles occupying superficial dermis, infiltrating into the deep dermis. High-power magnification showed a focal ulcerated area and a combined population. The first population was characterised by mild atypical eosinophilic epithelial cells. Admixed with the later, there was a second type of cells represented by pigmented epithelioid to stellated shaped within the same stroma. The tumour was confined to the basal membrane (*in situ*). There was no evidence of invasive malignancy.

The histological differential diagnosis in this case include melanoacanthoma, squamous cell carcinoma (SCC) colonised by atypical melanocytes, collision tumours of malignant melanoma (MM) and SCC, pigmented SCC, MM with adnexal extension (Figures 1 and 2).

Immunohistochemistry was performed to better identify

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Figure 1. *Hematoxylin and Eosin Stain (H&E) of a Heavily Pigmented Squamous Cell Proliferation at Low Power (x2)*

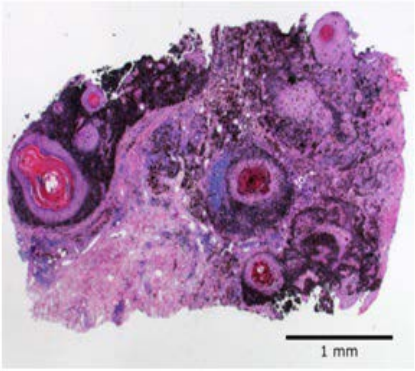


Figure 2. *A Higher Magnification (x 20) Showing the Intermingled Proliferation of Pigmented Cells (melanocytes) and Squamous Cells with Keratin Formation*

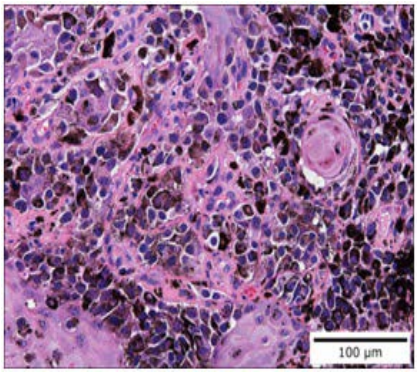


Figure 3. *Combined Immunohisto Chemistry: S100 (black) Highlighting Melanocytes Together with CK5/6 (red) Staining Squamous Cell with Keratin Formation*

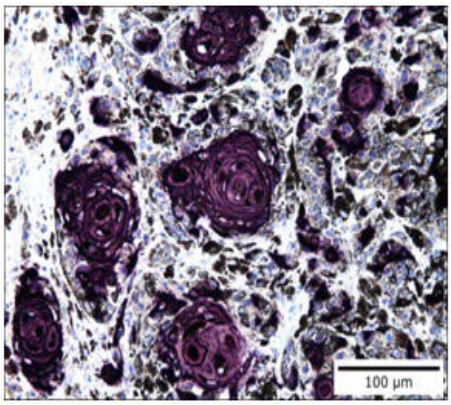
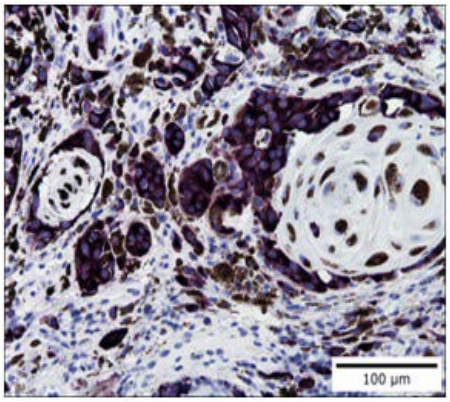


Figure 4. *Melan A Marker: Highlighting Melanocytes. Note that the Squamous Cells are Not Staining*



the nature of the cells. Figure 3 shows a combined immunohistochemistry. Due to the heavy brown pigment CK5/6 marker was performed in red colour showing cytoplasmic positivity for squamous cells. Together with that, there was a strong nuclear and cytoplasmic positivity for S100 (black colour), highlighting the melanocytes within the keratin formation. Another melanocytic marker (Melan A) was performed, showing strong cytoplasmic positivity which stained negative in the squamous cells (Figure 4). The presence of markers for both types of cells confirms the squamous and the melanocytic components, therefore supporting a diagnosis of a squamous-melanocytic tumour (SMT).

DISCUSSION

Squamous-melanocytic tumours are an uncommon and rare cutaneous neoplasia. There are only eighteen reported cases in the literature (Table 1) and none of them located in the anal canal. The squamous melanocytic lesion was first described by Rosen et al[6] and further restudied and classified as SMT by Pool et al.[8]

A literature review by Pool et al,[8] Satter et al[11] and Wang et al[3] who had evaluated tumour sites, age, gender and metastasis follow-up can be seen in the table below (Table 1). The great majority of those tumours are located in the head and neck region[3,8,10,12-14] and just one case was showed metastasis.[4] No gender predilection were seen as reported by Boyd et al[17] and the average age varied from 30s to 90s according to Wang et al.[3]

The theories for the tumoural histogenesis have been debated and there are four believed categories[8,12,14]: colonization tumour, combined tumour, collision tumour and biphenotypic tumour. Our case can be placed on the combined tumour classification.

Overall, we have presented a rare tumour with unusual location, since the most common location is head and neck and yet none of them occurred on the anal region. Although histogenesis remains unclear,[5,14] some papers support sun exposure and scarring process as a main tumorigenesis factor.[7] Our findings go against the sun exposure theory in as much as anal region usually is not a sun exposed area.

To sum up, we reported a rare neoplasia with dual component, composed of both malignant squamous and malignant melanocytic cells, known as squamous-melanocytic neoplasia. This case is the first to report the lesion on the area and is important to illustrate and to alert how it can easily be mistaken by its dif-

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Table 1. *SMT Location and Outcome in the Literature*

SMT	Age	Gender	Location	Metastasis
Muhlemann et al[5]	59	M	Trunk	Unknown
Rosen et al[6]	Unknown	Unknown	Unknown	Unknown
Walker and Walker[7]	78	F	Tigh	Unknown
Akiyama et al[4]	55	M	Right lower leg	Yes
Pool et al[8]	44	M	Head & Neck	No
	47	F	Head & Neck	No
	50	M	Head & Neck	No
	70	M	Head & Neck	No
Cutlan et al[9]	72	F	Shoulder	No
Dorić et al[10]	61	F	Head& Neck	No
Satter et al[11]	63	F	Left leg	No
	73	F	Left forearm	No
Rongioletti et al[12]	94	M	Back	No3
Pouryazdanparast et al[13]	62	M	Head & Neck	No
Leonard et al[14]	68	M	Head & Neck	No
Miteva et al[15]	82	M	Head & Neck	No
Amerio et al[16]	32	F	Right arm	No
Wang et al[3]	63	F	Head & Neck	No
Present study	60	M	Anal canal	No

SMT: Squamous-melanocytic tumour; M: Male; F: Female

ferentials. This is a neoplasia with uncertain biological potential and prognosis, with treatment relying on complete excision and observation. Due to the uncertain behaviour of these lesions, close follow-up is strongly advised.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

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