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Visceral Malignant Melanoma: Wilderness of a Black Hole

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Abstract **Case Report**

Malignant melanoma (MM) is malignancy of melanocytes, with the commonest primary site being skin. Mucosal melanomas make less than 1% and prognosis is poor if not detected early. Identifying the primary lesion can be challenging and a small percentage of patients falls under the category of melanoma of unknown primary (MUP). We herein report a rare case of an 86- year- old male who presented with frank hematuria and multiple pigmented naevi over bilateral lower limbs. A Computed Tomography Urography (CTU) revealed a bladder tumour at lateral wall, which then proceeded with a transurethral resection of bladder tumour (TURBT). He was discharged well pending histopathological examination (HPE) report. He presented again with small bowel obstruction and a CT showed small bowel intussuscepting tumour with several lungs and right adrenal gland lesions requiring a laparotomy, small bowel resection and anastomosis. A black pigmented tumour was found to be a lead point of the intussusception. Patient recovered well from surgery and was discharged well. HPEs of both the surgeries revealed malignant melanoma, unable to identify the primary tumour. This case shows the notorious behaviour of MM to metastasize without symptoms. Coupled with the similar HPE of both sites of bladder and bowel, it makes it difficult to establish the primary site. CT imaging should be the mainstay choice of investigation to identify primary disease and evidence of metastasis. Initial management of MM should be complete surgical resection of suspected primary lesion. In the unfortunate event of patient developing distant metastasis, role of adjuvant therapy in visceral melanoma is evolving with clinical trials focusing on immunotherapy, tyrosine kinase inhibitors and dacarbazine.

Keywords: Malignant melanoma, visceral, melanoma of unknown primary, dacarbazine, immunotherapy.

INTRODUCTION

Malignant melanoma (MM) or simply melanoma, is a malignancy of melanocytes, localized primarily on skin, accounting up to 95% of all melanomas [1]. Mucosal melanomas account for less than 1% of all malignant melanomas [2]. It is a serious disease, the prognosis of which is poor if not detected and treated early [3]. Most cases of malignant melanoma are diagnosed at an early stage when surgical excision can be curative. However, some patients have metastatic disease at presentation or develop metastases after their initial definitive treatment [4]. Although the vast majority of melanomas have a known primary site, approximately 3.2% of all melanomas present in distant sites with no known primary site, termed melanoma of unknown primary (MUP) [5] MUP are usually are found in lymph nodes, subcutaneous and then least commonly in visceral organs. De novo formation is current the debatable topic of the primary origin.

CASE REPORT

An 86-year-old male with well controlled hypertension and benign prostatic hyperplasia presented with painless frank hematuria for two-month duration. He was otherwise stable haemodynamically, no coagulopathy with a haemoglobin of 11.2. He was put on urinary bladder irrigation and an ultrasonography of the kidneys, ureters and bladder revealed an irregular mass. We proceeded with a Computed Tomography Urography (CTU) which revealed a suspicious urinary bladder tumour measuring 4.4cm x 3.3cm x 2.6cm located at lateral wall. He underwent transurethral resection of the bladder tumour (TURBT) and was discharged well two days after the surgery. On day five post-operatively, he presented again to the emergency unit symptoms of intestinal obstruction. Abdominal examination revealed a distended abdomen with active bowel sounds. No signs of peritonism. He was noted to have multiple naevi over bilateral lower limbs, however they were not suspicious in nature for a cutaneous

melanoma. A Computed Tomography (CT) of abdomen and pelvis showed small bowel obstruction secondary to intussusception likely due to a soft tissue tumour, and there were several lungs and right adrenal gland lesions suggestive of metastasis. He underwent an emergent laparotomy which revealed an intussuscepting pigmented tumour, 200cm from the duodeno-jejunal junction. Small bowels proximal to it were dilated with collapsed distal bowel segments. No others lesions found intraperitoneally. 50cm of small bowel was resected with a primary end to end anastomosis. He was discharged on day 5, once feeding was established. Histopathological examination (HPE) of the bladder and small intestine tumours were malignant melanoma, however unable to point out the primary lesion. He was counselled for adjuvant therapy, however he refused. He remained well during follow up with no new complaints.

Figure 1: (a-b) Physical examination of left lower limb showing multiple naevi

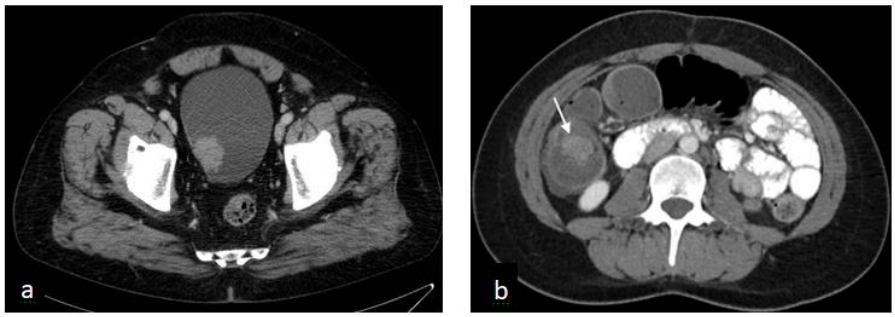

Figure 2: (a) CTU showing a tumour in lateral wall of urinary bladder. (b) Contrasted CT abdomen and pelvis showing target sign in small intestine, pathognomonic for intussusception.

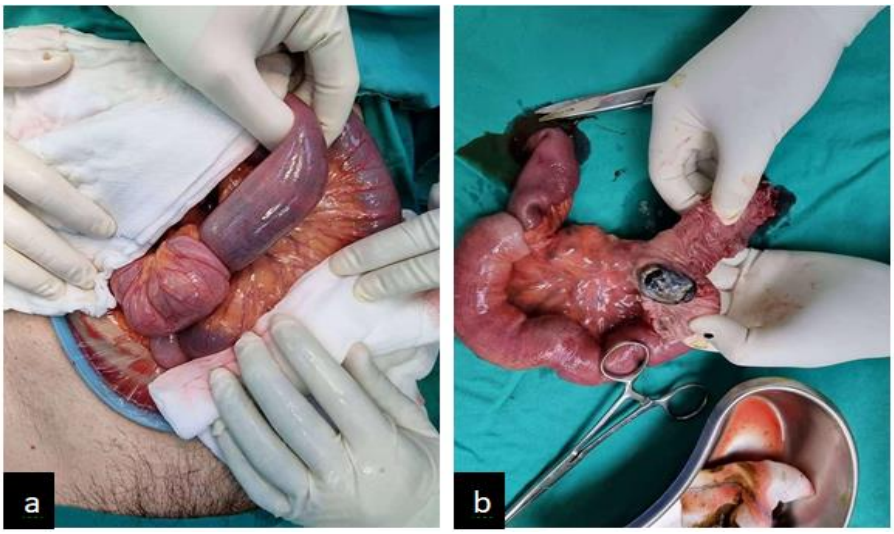

Figure 3: Intraoperative findings: (a) Small bowel intussusception with congestion. (b) Bi-valved resected small bowel specimen containing a black colored tumour arising from the mucosa

DISCUSSION

Malignant melanoma is notoriously known for its ability to metastasize. Identification of the primary of metastatic malignant melanoma is a difficult task till date. It could be the lack of published data, and also due to the poorly understood behaviour of the disease. The histopathology of each site, does not largely differ from one another which makes it even more difficult [6].

CT imaging is the mainstay of choice to identify the primary of this disease, where by the knowledge in recognizing the pattern and usual primary site of the disease is important [7].

As per retrospective study done by T Trout *et al*., in 2013, based on CT findings, it was found that the head/neck is the commonest site for primary, followed by back, lower extremities and the anterior chest wall [8].

Primary site	**Cases with atypical metastases n (% of cases with metastases)**	**Cases with only atypical metastasis**			
		N	**% of cases of that primary**	**% of cases of that primary**	**% of cases with only with metastases atypical metastases**
Head/neck	14/24 (58.3)	4	4.5	16.7%	19.0%
Back	20/40 (50)	5	4.4	12.5%	23.8%
Lower extremity	15/54 (27.8)	6	5.8	11.1%	28.6%
Anterior chest wall	5/12 (41.7)	2	6.1	16.7%	9.5%
Ocular	5/20 (25)	0	0	0%	0%
Upper extremity	4/8 (50)	1	3.4	12.5%	4.8%
Pelvis	2/5 (40)	1	6.7	20%	4.8%
Anorectal	1/3 (33.3)	0	0	0%	0%
Sinonasal	1/3 (33.3)	0	0	0%	0%
Anterior abdominal wall	0/1 (0)	0	0	0%	0%
Labia/vagina	0/1 (0)	0	0	0%	0%
Oral mucosa	0/0 (0)	0	0	0%	0%
Enteric	0/0 (0)	0	0	0%	0%
Unknown	4/10 (40)	2	13.3	20%	0%

In lieu with the lack of large sample size, studies now can only detail down the locations and possible common site of the metastasis to map out a possible pattern. More pathologic and clinical sampling are needed to aid to better understand the disease.

A study also suggest that a primary lesion may originate from the lymph node from a benign melanocyte, and also another study suggest that cutaneous melanoma may have been destroyed from trauma or the regression of the melanocyte from the body immunity, makes it difficult to identify. Lesion may also be in a hidden location or too small to be identified.

It is suggested that immunity may have a role in the spontaneous regression of a melanoma. A further better understanding of this disease is necessary to understand the pattern and nature of this disease.

Regardless of the primary site, initial management for loco-regionally confined or complicated visceral melanoma is surgical resection. Complete surgical resection, if feasible, offers the best chance for prolonged disease- free survival. However, most patients will ultimately develop distant metastases despite adequate local disease control of the primary disease with surgery. Role of adjuvant therapy in visceral melanoma is evolving with clinical trials focusing on immunotherapy, tyrosine kinase inhibitors and dacarbazine.

CONCLUSION

Malignant melanoma has a predilection of atypical metastases which makes identifying the primary difficult even with CT imaging. However, CT imaging still remains the main choice of modality.

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