PRIMARY ANORECTAL MELANOMA: REPORT OF A CASE

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Primary anorectal melanoma is a rare, but aggressively malignant tumor. The most common manifestation is lower gastrointestinal bleeding. We report a case of a 70-year-old female with cirrhosis of liver who presented with hematochezia, and on Colonoscopy was found to have a brown-grayish protruding anorectal mass that proven to be a melanoma. A series of imaging studies revealed no evidence of extra-luminal invasion, lymph node metastasis, and distant dissemination of tumor. Thereafter, the tumor was removed by trans-anal excision. The patient remained well without any evidence of tumor recurrence during a 30-month follow-up. We suggest that primary anorectal melanoma should be suspected in patients with concomitant presence of lower gastrointestinal bleeding and pigmented anorectal tumor. In order to preserve quality of life, and lessen operative morbidity, a conservative local excision might be a treatment option for primary anorectal melanoma, particularly in debilitated patients.

Key words: anorectum, melanoma

INTRODUCTION

Primary anorectal melanoma is a rare but deadly tumor. It accounts for only 0.2% to 3% of all patients with malignant melanomas [1-3] and 0.1% to 4.6% of all patients with anorectal malignant tumors [4-6]. Reviewing the literature, there were 457 cases reported from 1857 to 1981 [7] and the first reported case was in 1857 [8]. The most common symptom of anorectal melanoma is lower gastrointestinal bleeding [1]. The correct diagnosis is often considered late, and symptoms are often present for several months before diagnosis. Surgical interventions either by local excision or by abdominoperineal resection are the treatment options for anorectal melanoma. The prognosis is dismal with a grave five-year survival rate of about 6% to 17% [1,4,7,9-13] and median overall survival period of 12 to 25 months [3,7,12,14]. We report a female patient of primary anorectal melanoma presented with hematochezia. Patient received local excision and remained well without evidence of tumor recurrence during a 30-month post-operative follow-up.

CASE REPORT

A 70-year-old female patient with cirrhosis of liver presented with hematochezia. The same symptom had occurred about one year ago. However, she did not seek any medical aid until this episode of bleeding. On arrival, the physical examination disclosed pale conjunctiva, pulse rate of 117/min., and blood pressure
of 120/70 mmHg. The examinations of the chest, heart, and abdomen were unremarkable. No pigmented skin tumor was found. A mass with uneven surface and firm consistence was palpable by digital examination at 3 cm above the anal verge. Hematological studies revealed hemoglobin 9.4 g/dL, platelet count 67 x 10^9/UL, and prolonged prothrombin time that was 3.6 seconds longer than that of control. The serum biochemical tests showed mild elevation in total bilirubin level of 1.9 mg/dL (normal: 0.2-1.4 mg/dL) and low albumin level of 2.6 g/dL (normal: 3-5 g/dL). Colonoscopy showed a brown-grayish protruding mass with irregular surface, approximately 3 cm in size and located at 3 cm above the anal verge (Fig. 1). Pathological examination disclosed infiltrative nests among the lamina propria of anorectal mucosa, comprising large polygonal or spindle cells containing melanin pigments (Fig. 2A). Immunostain for anti-HMB-45 antibody revealed strong positivity (Fig. 2B). Lower gastrointestinal series revealed a 3 cm mass lesion at anorectal area (Fig. 3A).

Abdominal and pelvic computed tomography showed an anorectal mass, but without any paraaortic, iliac or inguinal lymph node involvement (Fig. 3B). Thereafter, patient received trans-anal wide excision under the preoperative diagnosis of stage I anorectal melanoma. The excised specimen showed a 3x5 cm pigmented mass with firm consistence (Fig. 2C), and the tumor cells were involved into submucosal layer, but the margin was free. The post-operative course was uneventful. Patient remained well without any evidence of tumor recurrence during a 30-month outpatient follow-up.

**DISCUSSION**

Primary anorectal melanoma is a rare but malignant tumor that is usually detected late. The average duration before diagnosis is 5 months [1]. It accounts for approximately 0.2-3% of all melanomas [1-3] and 0.1-4.6% of anorectal malignant tumors [4-6]. The anorectal region is the third most common primary site of melanoma and is also the most frequent.
Fig. 2A. The histology of biopsy specimen showed infiltrative nests among the lamina propria of anorectal mucosa with evident melanin pigmentation (H&E stain, × 200).

Fig. 2B. Immunostain for anti-HMB-45 antibody revealed intensive cytoplasmic positivity (DAB as chromogen, × 200).

Fig. 2C. Grossly, the excised specimen showed brownish pigmentation with irregular surface.
Anorectal melanoma occurs predominantly in females, with increasing incidence particularly in the older age group [1-4, 7-9-16]. The most common symptom of anorectal melanoma is lower gastrointestinal bleeding that is sometimes misdiagnosed as hemorrhoid. Other presentations include pelvic mass, abdominal pain, and obstipation [1]. The diagnosis of anorectal melanoma is based on physical examination, colonoscopy and pathological examination. The typical endoscopic finding is black pigmentation in a nodule with ulceration and friability [17,18]. However, one-third of melanomas are amelanotic [15]. Because of abundant vascular supply and lymphatic drainage, regional invasion (stage II) or distal metastasis (stage III) frequently occur during presentation and account for approximately one-fifth to two-thirds of patients [1,3,4,12,14,15]. They are usually manifested by a large pelvic mass, diffuse bilateral pulmonary nodules, or diffuse liver metastasis [14]. Therefore, once the diagnosis is made, image studies including chest X-ray and computed...
tomography of abdomen and pelvis are required for staging.

Since anorectal melanoma is not responsive to radiotherapy, and the role of chemotherapy and immunotherapy remains unclear, surgical resection is the choice of treatment in the majority of cases [19]. The methods of surgical treatment include local excision and abdominoperineal resection with or without lymphadenectomy [3,12-14]. There are extensive controversies regarding which type of surgical procedures should be performed. Abdominoperineal resection does not seem to offer any significant difference in the survival time, but it does have some effect in controlling the local recurrence as well as in extending the disease-free period [12]. However, in order to minimize the associated operative or post-operative morbidity and preserve the quality of life, local excision might be the preferred choice for initial treatment, particularly in debilitated patients. In contrast, abdominoperineal resection should be reserved for those patients with bulky local disease that cannot be removed by local excision alone and for salvage control in selected patients with local recurrence [12]. In our case, the patient received trans-anal mass excision without lymphadenectomy. Such surgical treatment seemed to be good enough to maintain her quality of life without any complications resulting from the surgery or tumor itself.

Primary anorectal melanoma is a rare disease in Taiwan, and a rare cause of lower gastrointestinal bleeding. It should be suspected in patients presented with hematochezia and pigmented anorectal tumors during colonoscopic examination. In order to preserve the quality of life for debilitated patients, local excision might be the choice of treatment for primary anorectal melanoma.

REFERENCES


原發性肛門直腸黑色素瘤：一病例報告

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原發性肛門直腸黑色素瘤是一種罕見，具侵犯性的惡性腫瘤。下消化道出血是最常見的表徵。在此我們提出一例報告：一位有肝硬化病史之七十歲女性因血便至醫院就診，大腸鏡檢查發現肛門邊緣以上3公分處有一3公分大小之灰黑色的腫塊，經病理診斷為黑色素瘤。系列檢查並無發現它處之轉移。隨後此病患接受局部腫瘤切除，術後恢復良好並追蹤至今已三十個月，並無腫瘤復發之情形。因此，若患者出現下消化道出血及大腸鏡檢查發現黑色的腫塊應懷疑原發性肛門直腸黑色素瘤。對於肝硬化或其它慢性病之病人，為保有良好之生活品質與減少術中或術後併發症，局部切除應為另一選擇之手術方法。

關鍵語：肛門直腸，黑色素瘤