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Case report

Abdominal perineal resection or wilde local excision in primary anorectal malignant melanoma. Case report and review

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HIGHLIGHTS

- Anorectal melanoma (ARM) is a systemic disease.
- Regardless how aggressive is ARM, no surgical treatment will truly change the outcome.
- If surgical teqchniques are available, patients should undergo to wide local excision.
- In case of recurrence, the abdominal resection should be considered as the best surgical treatment.

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ABSTRACT

Introduction: Primary anorectal malignant melanoma is a rare and aggressive tumor that carries a poor prognosis. Anorectal melanoma (ARM) is often misdiagnosed as hemorrhoids adenocarcinoma polips and rectal cancer. ARM spreads along sub-mucosal planes and is often to wide-spread for complete resection at time of diagnosis and almost all patients die because of metastases.
Presentation of the case: A 77-year-old male patient presented a history of recurrent rectal bleeding and whose histopathological diagnosis was melanoma.
Discussion: The treatment of choice remains controversial. Surgery with complete resection represents the typical treatment. However standard operative procedures related to the area of resection and lymph dissection have yet to be established. Abdominal perineal resection (APR) with or without bilateral inguinal lymphadenectomy or wide local excision (WLE) have been used to manage patients with ARM.
Conclusion: The higher serum levels of LDH and YKL-40 are suggestive for Anorectal Melanoma diagnosis. The decrease of these findings may be associated with good prognosis. The review of both APR and WLE options suggests no significant difference in survival among patients.

1. Introduction

Primary anorectal malignant melanoma (AMM) is a rare and aggressive tumor with poor prognosis. It represents 1–2% of all melanomas and it is the third most common form of melanoma after the skin and retina. Nonetheless this condition is known, any description of single case may give more useful information on this pathology.

The disease typically affects Caucasians, females, and patients between the fifth and eighth decade of life with a mean incidence of 64.3 yrs (58.1 yrs and 70.2 yrs) [1].

The symptoms such as elimination of mucus and blood through the anal canal, presence of anal pain, feeling of rectal fullness or incomplete evacuation, externalization of tumor, and changes in bowel habits, are common to other tumors in anorectal region.

Many treatments including surgery, chemotherapy and radiotherapy have been used, but AMM is frequently radiotherapy-resistant and shows a poor response to chemotherapy [2]. Whether surgical procedure is better than other treatments is still an issue, and also it is controversial whether abdominoperineal resection of the anorectum (APR) or Wilde local excision (WLE) of

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the tumor has better outcomes.

In this report, we present the case of a patient with anorectal melanoma, without lymph node or distance metastases who underwent laparoscopic abdominoperineal resection (APR) [3,4].

2. Case presentation

A 77-year-old man was admitted to the hospital complaining of 8-months history of painless rectal bleeding. A digital rectal examination revealed a hemorrhagic, soft mass of rectum, 5 cm from the anal verge. The findings of the remainder of the physical examination were within normal limits. At the time of admission and during the follow up the serum was analyzed for evaluation for biochemical and tumor markers [5]. After transanal polypectomy, with sufficient macroscopically negative margins, the histologic result was consistent with ulcerated malignant *epithelioid* melanoma, without BRAF V600 mutation, involving rectal mucosa and submucosa, with positive lateral surgical resection margin and unclear deep margin. At admission LDH and YKL-40 are respectively 987 IU/L and 852 μg/L (Table 1).

Clinical history for primary melanoma in the skin and in other noncutaneous sites, including the eye, was negative. A computed tomographic scan of the abdomen, chest, brain, and PET/CT (Positron Emission Tomography - Computed Tomography) showed a non homogeneous and partially calcified mass occupying the prostatic bed, and excluded lymph node and distant metastases.

A laparoscopic abdominal perineal resection with anorectal amputation was carried out, along with total mesorectal excision using curved harmoning shears Ultracision®.

The histologic examination of the operative specimen showed only a tumor to invade through the lamina propria of mucosa at anorectal junction. At histology, the case was diagnosed as stage I and T1b Breslow's thickness (Figs. 1 and 2).

One year after surgical intervention a full-body computed-tomography (CT) scans, LDH and YKL-40 excluded lymph node and distant metastases.

3. Discussion

Anorectal melanomas are rare but aggressive tumors. Compared with cutaneous melanomas, anorectal melanomas have the lowest percent of five years survivals, only 25%. Best hope for survival is offered by early detection and complete surgical removal. However is usually delayed because of occult site of recurrence and unspecific symptoms diagnosis. The most common presenting symptom of anorectal melanoma is bleeding with 53–89% of patients reporting this as the predominant complaint. Other symptoms are suspicion of hemorrhoids, discomfort or pain, an anal mass, change in bowel habit, tenesmus or pruritus. The higher serum levels of LDH and YKL-40 are suggestive for suspicion of anorectal melanoma diagnosis. A smaller proportion of patients present with nodal disease and inguinal lymphadenopathy. Due to absence of specific symptoms and comparing with other anorectal disease, anorectal melanoma is often diagnosed accidentally. Although rare, several cases have been described, each case gives information on both treatment and prognosis of AMM [6,7].

There are two methods of staging in anorectal melanoma: 1) developed by The American Joint Commission on Cancer, which is a staging method based on depth of primary tumor, presence of tumor in lymph nodes and presence of distant metastasis. 2) Another staging system based only on disease spread: it describes local disease only as stage 1, regional lymph node disease as stage 2, and metastatic disease as stage 3 [8].

Tumors are most commonly located in the anal canal, followed by at the dentate line with fewer melanomas located in the rectum itself [9].

There is no consensus on which surgical approach is preferred. A number of studies claim that abdomino-perineal resection (APR) is the treatment of choice [10] because it can control better lymphatic spread and it allow to obtain larger negative margins for local control.

Other studies instead have recommended only a sphincter-saving excision (wide local excision) because treatment is often palliative and wide radical surgery is unnecessary mutilating [11]. Wide local excision is defined as a sphincter-saving operation with a defined margin around the tumor in two dimensions. The benefits of a WLE are quicker recovery, no need for a stoma, and minimal

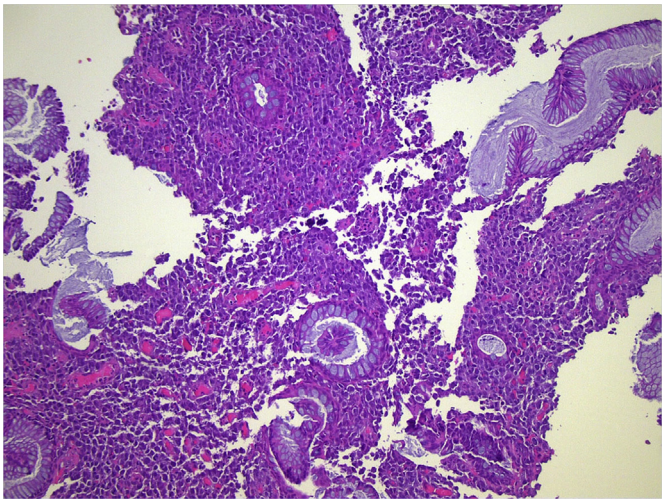

Fig. 1. Presence of melanoma in intestinal cells (hematoxylin and eosin staining).

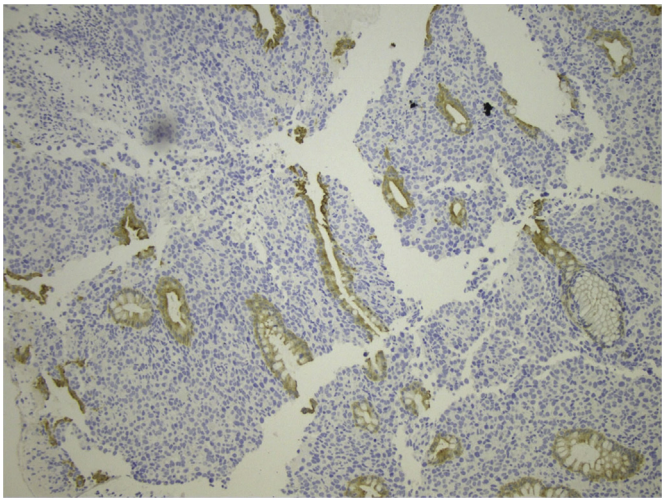

Fig. 2. Presence of cytokeratin (CKAE1/AE3) in intestinal cells.

Table 1
Serum markers of the patient.

	CEA	Ca 19-9	LDH	YKL-40
Normal value	0–5 IU/L	0.39 IU/ml	120–250 IU/L	45–500 μg/L
At admission	4.88	44	987	852
7 days after surgery	3.25	38	654	528
1 month after surgery	3.40	35	525	460
1 year after surgery	2.88	27	256	396