CASE REPORT

Primary malignant melanoma of uterine cervix with probable origin from benign cervical melanosis

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SUMMARY

We report a case of primary malignant melanoma of cervix which is a rare malignancy with only around 60 cases being reported. This patient presented with bleeding per vaginum. A large exophytic growth from cervix with black discolouration was seen. International Federation of Gynaecology and Obstetrics (FIGO) staging was stage IIa. Histoimmunocytology confirmed the diagnosis of malignant melanoma of cervix. S-100 and HMB-45 are reliable markers for cervical melanomas. We attempted Wertheim's hysterectomy; but, due to extensive disease, paravescical and paraarectal fossae could not be dissected and we had to perform type I hysterectomy. The patient was started on adjuvant chemotherapy with dacarbazine. Despite counselling, she refused a complete course of chemotherapy and died after 6 months. Radical surgery, chemotherapy and immunotherapy are the therapeutic modalities used. In bulky disease, neoadjuvant chemotherapy should be considered to reduce the tumour bulk and facilitate required surgery. Prognosis is poor and unpredictable.

BACKGROUND

Primary malignant melanoma of cervix is a rare entity and around 60 cases have been reported to date. Genital melanomas constitute 5% of melanocytic malignancies in women and cervical melanomas are most unusual among them.¹ We are reporting a case of primary malignant melanoma of cervix with grossly obvious lymph node metastasis, also confirmed by histopathological examination.

CASE PRESENTATION

A 35-year-old woman, belonging to low socioeconomic strata, presented to the gynaecology clinic presenting with brownish discharge per vaginum and irregular vaginal bleeding since 2 months. There was no history of recent growth, itching or pain in any mole over the body. General physical examination was normal. An extensive dermatological, mucosal or uveal examination did not reveal any other primary melanoma. On speculum examination cervix was replaced by black coloured exophytic growth occupying upper two-third of vagina (figure 1). Vaginal involvement was limited to its upper third, but there were black satellite lesions in lower vagina too (figure 2). On gynaecological examination, cervix was bulky, uterus was normal in size, bilateral adnexae were not palpable and parametrium not involved. Clinical diagnosis of International Federation of Gynaecology and Obstetrics (FIGO) stage IIa was carried out.

Figure 1 Image showing cervical melanoma as large, polypoidal mass filling upper third of vagina with black pigmentation.

INVESTIGATIONS

Cervical biopsy was taken and histological and immunohistochemical findings were suggestive of malignant melanoma of cervix. Microscopic examination of the tumour showed sheets of predominantly monomorphic cells, with few cells showing dark-brown pigment. The cells were positive for S-100 and HMB-45 and stained for Schmorl's stain. There was a presence of subtle epitheliotropism. Biopsies from satellite lesions in vagina revealed benign melanocytic proliferation. Blood investigations and chest x-ray were normal. MRI of pelvis showed bulky cervical growth. No other site of tumour involvement was seen in pelvis. Abdominal MRI was normal. We wanted to get a CT scan of the chest, but the patient could not afford any more investigations.

Figure 2 Image showing satellite lesions on posterior wall of vagina which on histopathological examination were benign melanosis.

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DIFFERENTIAL DIAGNOSIS

Melanosis of the uterine cervix is an incidental finding usually seen in postmenopausal patients with uterine prolapse. Early stages of malignant melanoma may look like benign melanosis. Amelanotic forms of malignant melanoma may require differentiation from the other common cervical malignancies. Rare tumours like cervical malignant peripheral nerve sheath tumour and sarcoma uterus have also been reported to resemble its amelanotic form.² Common blue naevus of the uterine cervix can also be considered to be one of the differentials.³ Cervical endometriosis may sometimes present as black lesions on cervix and at times it may also present as exophytic growth.⁴ Immunohistochemical study and histopathology is useful to differentiate these conditions from melanoma.

TREATMENT

Surgery has been described as the treatment of choice in any melanoma including urogenital melanomas.^{5–7} We attempted to perform Wertheim's hysterectomy for the patient, but we were not able to perform as the dissection of paravascular and pararectal fossae was not possible due to extensive disease (figure 3). We still proceeded with pelvic and para-aortic lymph node dissection and could identify black-coloured pelvic lymph nodes during dissection. A total of 18 lymph nodes (pelvic and para-aortic) were resected. Histopathology and immunohistochemistry of the specimen confirmed the diagnosis of malignant melanoma, and 15 of the 18 lymph nodes were also found to be positive for malignancy. She received one cycle of adjuvant chemotherapy with cisplatin, vinblastine and dacarbazine. The patient had severe episodes of vomiting during the course of chemotherapy which were controlled by antiemetics. The patient refused to receive further chemotherapy despite counselling. She was discharged on request from hospital.

Figure 3 Image showing hysterectomy specimen with large, black cervical tumour.

OUTCOME AND FOLLOW-UP

The patient did not follow-up with us; however, on enquiry, we found that she had died 6 months after discharge from hospital.

DISCUSSION

Malignant melanoma of the cervix is a very rare lesion and no prospective studies exist. According to a retrospective study⁶ comprising of four patients, the average age of patients was 45 years. Presenting symptoms constituted irregular vaginal bleeding, postcoital bleeding or vaginal discharge. Gynaecological examination showed polypoidal cauliflower-shaped or nodular black-brown or black-blue mass on the cervix. In another study,⁷ in which data were compiled from previous case reports, the patients' ages ranged from 39 to 78 years; the presenting symptom was vaginal bleeding in 67% cases; the majority of the patients (58%) presented in their early stages (Ia–IIa). On literature review, we found that the age of patients varied from 19 to 78 years. The most common presenting complaint was vaginal bleeding (72% patients). On clinical examination, the appearance of lesions varied from black-brown, blue-brown or dark-coloured ulcerative or polypoidal growth. At times, they could not be differentiated from other cervical malignancies. In one of the cases, it was diagnosed after a routine Pap smear in asymptomatic woman which showed high-grade squamous intraepithelial lesion.⁸ Majority of patients (86%) presented in stage I or II. Eighty per cent of the reported cases of cervical melanoma are primary tumours and the rest are metastatic.

Primary malignant melanoma either originate from melanocytic elements within epithelium or from cervical stroma.⁹ In our patient, the possibility of origin of malignant melanoma from benign melanosis over cervix is present as there were benign melanotic spots over posterior vaginal wall. Cervical melanoma is diagnosed by histopathology and confirmed by special staining procedures which reveal layers of monomorphic cells with interspersed melanin-containing cells. Schmorl's or Masson's Fontana stain is used for melanin.¹⁰ S-100 and HMB-45 were positive in all previously reported patients of cervical melanoma, and immunocytochemical staining with these markers plays a very important role in the diagnosis of primary malignant melanoma of cervix. These markers were positive even in the clear cell variant of cervical melanoma. Diagnosis by cervical fine-needle aspiration cytology and scrape cytology has also been reported which showed bizarre and abnormal cells containing pigment.^{11 12}

The treatment of genital melanocytic malignancies as described in the literature is surgery combined with chemotherapy and immunotherapy in early stages, which is the preferred therapeutic approach. Surgery in cases of cervical melanomas ranges from simple excision to radical hysterectomy or Wertheim-Meigs hysterectomy with bilateral adnexectomy and regional lymphadenectomy. Treatment controversies exist and some recommend lymphadenectomy only for grossly positive nodes.⁷ Chemotherapy with immunotherapy is the recommended treatment in advanced cases. The chemotherapeutic agents used in the treatment of melanoma are dacarbazine, cisplatin, interleukin (IL)-2 and Vinblastine.¹ Chemotherapy in combination with dimethyl triazemo imidazole carboxamide and biological therapies with interferon- γ and IL-2 have also reported to improve the prognosis of the primary malignant melanoma of cervix in early stages.⁶ Radiotherapy has also been advocated by some and in one of the case reports the patient received adjuvant pelvic radiation therapy with a total dose of 4500 cGy.⁷

Complete remission of malignancy has been achieved with combined radical surgery and irradiation in few reported cases.¹³ The survival of patients in FIGO stage II after radical surgery and adjuvant chemotherapy ranges between 6 months and 7 years as reported in previous cases.

No consensus regarding treatment modality of choice for this tumour exists due to its rare nature. Similar to malignant melanoma at any other site, surgery can be considered the mainstay of treatment as these tumours are relatively chemoresistant. Surgery has been reported for the treatment of cervical malignant melanoma even in bulky tumours unlike squamous cell carcinoma or adenocarcinoma of cervix when chemoradiation is preferred treatment.¹⁴ In our patient, we tried to perform Wertheim's hysterectomy, but could not succeed due to extensive disease. We feel that neoadjuvant chemotherapy with dacarbazine could have benefitted and made the appropriate surgery feasible.

Learning points

- ▶ Presenting symptoms can be irregular vaginal bleeding, postcoital bleeding or vaginal discharge. Examination may reveal polypoidal cauliflower-shaped or nodular black-brown or black-blue mass on the cervix.
- ▶ S-100 and HMB-45 are reported to be positive in all previously reported patients of cervical melanoma and immunocytochemical staining with these markers play very important roles in the diagnosis of primary malignant melanoma of cervix
- ▶ The prognosis of cervical melanomas is poor and unpredictable. Widespread survival has been reported within the same stage.
- ▶ Though no treatment guidelines exist due to rare nature of tumour, complete remission of malignancy has been reported to be achieved with combined radical surgery and irradiation or adjuvant chemotherapy.
- ▶ In bulky cervical tumours, neoadjuvant chemotherapy can be considered as it can make the required extensive surgery more feasible.

Competing interests None.

Patient consent Obtained.

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