Case Report

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Plasmacytoma of the cervix

Katherine Marie A. Tuason, Concepcion D. Rayel

Abstract:

Plasmacytoma is a rare disease entity that represents only 5%–10% of all plasma cell neoplasms. It is rarely found in the female genital tract. There are 8 identified case reports on plasmacytoma of the cervix. The clinical symptoms are nonspecific and biopsy with immunohistochemistry is used to diagnose. Due to the paucity of cases, there is no standard treatment modality. We present a case of a 34-year-old patient who had a history of postcoital bleeding. Biopsy and immunohistochemistry were done which confirmed the diagnosis of plasmacytoma. Total abdominal hysterectomy, bilateral salpingectomy with transposition of ovaries was the chosen treatment option.

Keywords:

Extramedullary plasmacytoma, female genital tract, solitary plasmacytoma, uterine cervix

Introduction

Plasmacytoma is an abnormal proliferation of monoclonal plasma cells. It is a lesion that can either be located in the bone or in other areas of soft tissues. It is a rare disease entity that represents only 5%–10% of all plasma cell neoplasms. This generally occurs more in men than women with incidence rates 2–3 times higher. It mainly affects people in the 5th to 6th decade of their lives, and the risk increases in people over the age of 60. It also affects African Americans more than Caucasians, with the lowest incidence reported in the Asian population.[1-4]

The disease exists in two forms: solitary bone plasmacytoma (SBP) and solitary extramedullary plasmacytoma (SEP) . The incidence of SBP is approximately 40% more than that of SEP.[3] A SBP arises mostly from the axial skeleton and while a solitary extramedullary plasmacytoma mostly arises from the head-and-neck region particularly affecting the respiratory tract.[1-4] These lesions are rarely found in the female genital tract.

This case is a rare occurrence of plasmacytoma in the uterine cervix. It presents with symptoms of vaginal bleeding, abnormal pap smear findings and a suspicious looking cervix. Because of the rarity of this disease, the optimal treatment method is not established. Based on literature, treatment recommendations include radiation therapy or surgery for resectable lesions.

Case Report

A 34-year-old, G2P2 (2002), married, Filipino, consulted for an episode of postcoital bleeding. She had no known comobidties and had previously delivered vaginally and abdominally. She is a housewife, a nonsmoker, and nonalcoholic beverage drinker. She had her menarche at 12 years of age with subsequent menses occurring at regular intervals, lasting 3–5 days using 3–4 pads/day with associated dysmenorrhea on the 1st day. She was on intermittent oral contraceptive pills for 9 years and she had no history of sexually transmitted infections.

A pap smear was done and the result showed atypical squamous cells of undetermined

Department of Obstetrics and Gynecology, Davao Doctors Hospital, Davao City, Davao del Sur, Philippines

Address for correspondence:
Dr. Katherine Marie A. Tuason,
118 Elpidio Quirino Ave, Poblacion District, Dvavao City, 8000 Davao del Sur
E-mail: kattuason@gmail.com

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significance. With a suspicious-looking cervix, [Figure 1] she was referred to a specialist for colposcopy and biopsy. Acetowhitening was noted and biopsy of the cervix with endocervical curettage were done. The histopathology report revealed a proliferation of atypical lymphoid cells. With these findings, a simple reactive process or a malignant lymphoma was considered. The pressing need to rule out a malignant process prompted further investigation by doing immunohistochemistry. A lymphoma panel was requested which included T-cell markers: CD3 and CD4 and B-cell markers: CD19 and CD20. The B-cell marker, CD19, was strongly positive which indicated the presence of immature lymphocytes and plasma cells. Further tests were done to identify the origin of the problem. A positive expression of CD138 and MUM1 confirmed a plasma cell origin [Figures 2 and 3].

A plasmacytoma of the uterine cervix was considered. This diagnosis was supported by the demonstration of monoclonal plasma cell infiltrate without evidence of myeloma at any other site. Hence, a biopsy of the bone marrow aspirate, a peripheral blood smear, and other blood chemistries were done to investigate a systemic problem. The normal results of the aforementioned tests and the presence of monoclonality expressed by kappa light chain restriction [Figure 4] clinched the diagnosis of plasmacytoma.

She underwent total abdominal hysterectomy, bilateral salpingectomy with transposition of ovaries. The biopsy result of the specimen revealed residual atypical plasma cells at the cervix located in the 9'o clock position, measuring 0.4 cm and the rest of the specimen was normal.

Discussion

A plasma cell neoplasm is an abnormal proliferation of monoclonal plasma cells that can either present as a systemic condition or as a solitary lesion. The most common disease entity is multiple myeloma, which affects the bone marrow throughout the axial and appendicular skeleton. Its manifestations include bone pains and pathologic fractures. Diagnostic tests would reveal anemia, hypercalcemia, presence of renal insufficiency, bone marrow plasmacytosis of more than 30%, and lytic bone lesions on skeletal survey.[1,3,5]

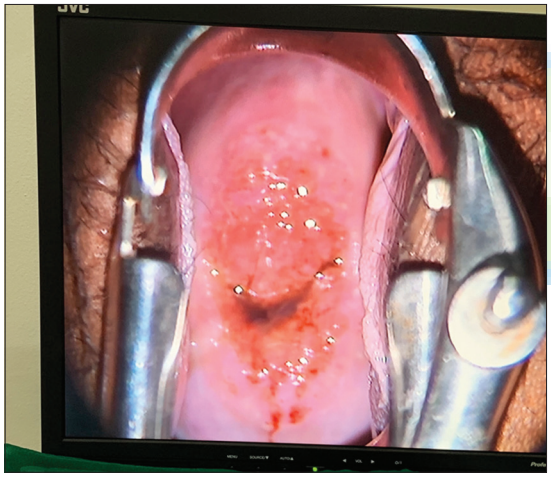

Figure 1: Cervix

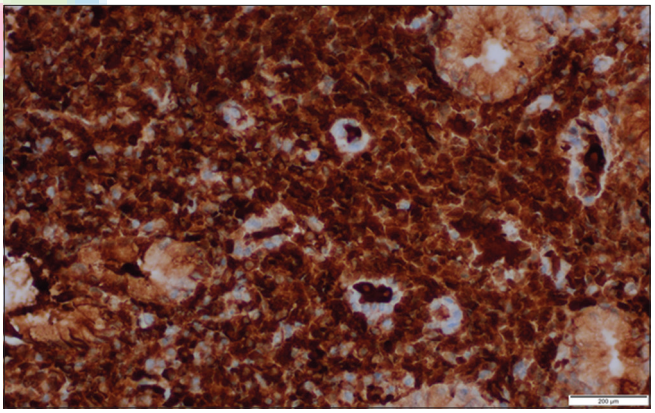

Figure 2: Cervix. Expression of kappa immunoglobulin light chain

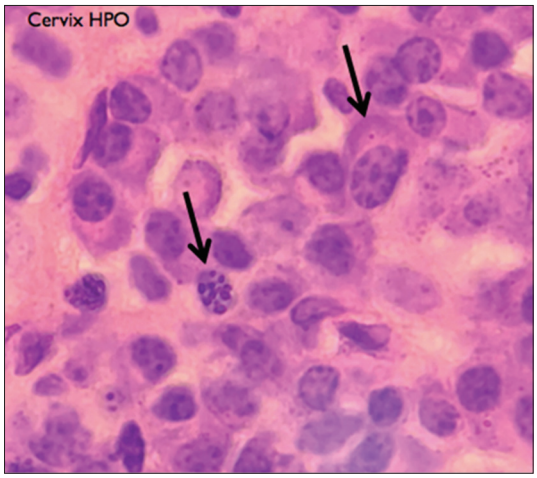

Figure 3: Cervix. Atypical plasma cells

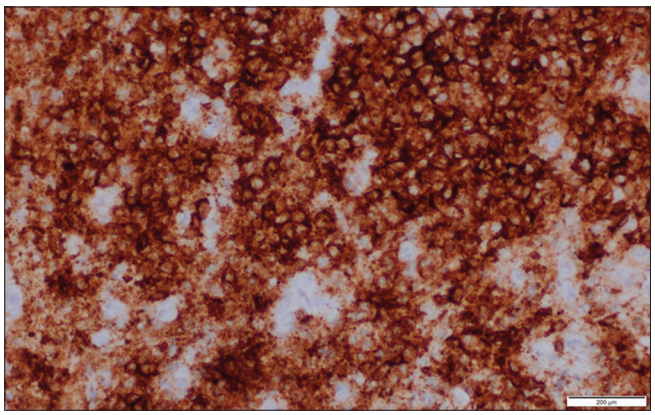

Figure 4: Cervix. CD138 positive

A solitary plasma cell lesion is called a plasmacytoma, which is a rare neoplasm. It is characterized by a discrete, solitary mass of neoplastic monoclonal plasma cells. Although it does not present with systemic problems, it has been unequivocally identified as a risk factor for progression to multiple myeloma. The 5-year risk of progression is 30%–50% for patients presenting with SBP and 10%–30% for those presenting with SEP.[3,5] Rowell *et al*. enumerated the diagnostic criteria for solitary plasmacytoma: (1) a biopsy proven, single infiltrate of clonal plasma cells in the bone (SBP) or soft tissue (SEP); (2) a bone marrow biopsy showing no evidence of infiltration by clonal plasma cells; (3) a negative skeletal survey and; and (4) the lack of systemic symptoms that would suggest multiple myeloma. These diagnostic criteria are similar to that of Rosai *et al*. and Grammatico *et al*.[6]

There is no established identifiable cause of plasmacytoma. Certain factors such as viral infections, prolonged irritation, and genetic factors play a role.[1,2] The symptoms of plasmacytoma are often nonspecific and vary depending on the site. The most common symptom is pain.[1]

The presence of a solitary plasmacytoma in the female reproductive tract is a rare occurrence. There are case reports of ovarian, cervical, and vaginal plasmacytoma. Based on the comprehensive study done by Feldman *et al*., there are 11 cases of ovarian plasmacytoma, 8 cases of cervical plasmacytoma, and 5 cases of vaginal plasmacytoma. They presented with signs and symptoms varying from a palpable abdominal mass, decreased appetite, weight loss, and irregular vaginal bleeding to postcoital bleeding.[7,8]

In the case presented, postcoital bleeding was the primary and only symptom. The suspicious looking cervix prompted further investigation with the use of colposcopy and cervical biopsy, which revealed atypical lymphoid proliferation. A plasma cell typically shows a round-to-oval eccentric nucleus, coarse and condensed chromatic with basophilic abundant cytoplasm, and a perinuclear halo. However, in this case, atypical plasma cells were noted in the cervical biopsy, which showed eccentric nuclei, abundant cytoplasm, mild-to-moderate nuclear atypia with hyperchromatic nuclei, and clumped chromatin [Figure 5]. With the histologic presence of plasma cells, it is important to exclude a reactive process or other neoplastic processes mainly multiple myeloma and other plasma cell neoplasms.[2,9] The biopsy specimen of the cervix was subjected to immunohistochemistry. The B-cell marker CD19 was identified to be strongly positive. Further immunohistochemistry revealed that CD138 and MUM-1 were positive, confirming the plasma cell histogenesis of the neoplasm. Laboratory workup did not show any signs of systemic manifestations of multiple myeloma. Bone marrow aspirate biopsy was done which revealed results of a normocellular marrow, negative for atypical cells.

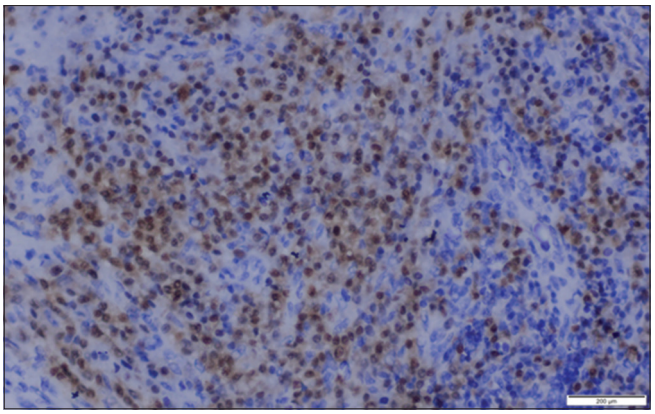

Figure 5: Cervix. MUM1 positive

To strengthen the diagnosis of extramedullary plasmacytoma, the demonstration of a monoclonal plasma cell infiltrate proliferation is needed.[5] Monoclonality needs to be proven by kappa or lambda light chain restriction or by a polymerase chain reaction‐based approach. In this case, monoclonality is expressed by kappa light chain restriction with the absence of lambda light chain. The gathered morphologic and immunohistochemistry findings and the lack of evidence of myeloma at any other site confirm a diagnosis of plasmacytoma.[5]

Monoclonality needs to be proven by kappa or lambda light chain restriction or by a polymerase chain reaction-based approach. In this case, monoclonality is expressed by kappa light chain restriction with the absence of lambda light chain. The gathered morphologic and immunohistochemistry findings and the lack of evidence of myeloma at any other site confirm a diagnosis of plasmacytoma.

Due to the rarity of this disease, there is no standard treatment approach. Treatment modalities are primarily based on previous review of literature. Plasmacytoma is a highly radiosensitive neoplasm thus radiotherapy alone is recommended.[1,7,10] However, with lesions that can be completely resected, surgery alone can be considered as another option. It is unclear whether additional treatment with radiotherapy is necessary for resected lesions with clear surgical margins.[3] Adjuvant radiotherapy is done in some cases when complete resection cannot be achieved. In this case of a cervical plasmacytoma, hysterectomy is the most commonly used treatment approach.[2] Our patient underwent a total abdominal hysterectomy, bilateral salpingectomy with transposition of both ovaries. With eight cases of cervical plasmacytoma reviewed by Feldman *et al*.,

four were surgically treated and three out of four were followed up and identified to have no evidence of disease or progression of disease after a few months to years.

Indeed, there is paucity of data on plasmacytoma of the cervix. In general, patients treated for plasmacytoma are monitored every 3 months for the 1st 2 years, every 6 months for the next 3 years, and then, yearly for the next 5 years. During follow‑up, laboratory tests include complete blood count, serum creatinine, serum calcium, urine and serum electrophoresis. Additional imaging studies could also be requested preferably a positron emission tomography/computed tomography, magnetic resonance imaging or whichever is the imaging modality of choice on initial assessment. In this case, it is important to follow-up and continue surveillance, especially with the possibility of progression to multiple myeloma.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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