Case Report
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Malignant Mixed Mullerian Tumor of the Uterine Cervix: A Case Report

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Abstract

Malignant mixed mullerian tumors (MMMT) are infrequent and highly malignant tumors. MMMTs, emerging from the uterine cervix, are very rare and to the best of our knowledge, there are no specific symptoms for the diagnosis of MMMTs whose early diagnosis is challenging. Almost all of them are diagnosed with pathological tests and reports. A 52-year-old post-menopause woman was referred to us, who suffered from postmenopausal bleeding from four months earlier. Upon pelvic examination, the position of the biopsy was identified in the anterior lip of the cervix. In former fractional dilatation and curettage, we found a pathology report with MMMT in the anterior lip of cervix. We performed a radical hysterectomy type II. In the permanent pathological report, MMMTs stage IBI was established. The patient was followed by chemoradiation. After 20 months, examination showed no evidence of recurrence.

Keywords: Mixed tumor, Mullerian, Malignant neoplasm, Uterine cervical cancer, Gynecology

Introduction

Malignant mixed mullerian tumors (MMMT) are scarce and highly malignant tumors that emerge from the female genital tract.[1] MMMTs are biphasic, with two portions of carcinoma and sarcoma. The most common components are squamocellular carcinoma combined with homologous sarcoma.[2] The typical primary site of MMMTs is the uterine corpus and it is uncommon for MMMTs to develop in the uterine cervix, which may be only 0.005% of all cervical malignancies. To the best of our knowledge, until 2013, only 50 documented cases had been reported.[3,4] MMMTs are more frequently seen in postmenopausal women and usually present with abnormal vaginal bleeding.[3] There

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are no specific clinical manifestations for MMMTs and almost all cases are diagnosed by histological examination. In this article, we reported a case of MMMTs arising from the uterine cervix with stage IBI.

Case Presentation

A 52-year-old post-menopause woman was referred to our obstetrics and gynecology center in Mashhad, Iran, Ghaem Hospital. She suffered from postmenopausal bleeding from four months before (from July 2019). Upon pelvic examination, the site of the biopsy was identified in the anterior lip of the cervix and the clinical stage was IBI. She underwent fractional dilatation and curettage in August. In her documents, we found normal ultrasonography of uterine and ovaries in addition to a pathological report of MMMT in the anterior lip of cervix. The report indicated that in the cross-sectional study of the cervix, there were neoplastic glandular proliferation, hyperchromatic nuclei with atypia and polymorphism, and mixed stroma with cartilage focus in it. Paraclinical evaluations, such as ultrasonography and magnetic resonance imaging (MRI) with IV contrast, revealed that the only abnormal finding was increased enhancement of cervix that referred to tumoral lesion or inflammation. Therefore, in October 2019, the patient underwent radical hysterectomy type II. Pelvic and para-aortic lymphadenectomy was done. Subsequently, the patient underwent 25 sessions of external radiotherapy, three sessions of brachytherapy, and six courses of chemotherapy, including taxol with carboplatin. The patient has been fallowed every 3 months and after 20 months, examination showed no evidence of recurrence. All the physical examinations and imaging were normal.

Pathological findings

The cervix was 3.5 cm in length and exocervix was Oyster-color with a brown focal lesion in it with 4.3 × 3.3 cm in length. Once the pathologist cut it, a nodular well-defined lesion, 1 cm in diameter, was observed. Uterine and ovaries were in a normal shape and size. In microscopic investigations, MMMT at the right superior quadrant of the cervix (11 o'clock), 1cm × 1cm in size, was reported (Figure 1). All the

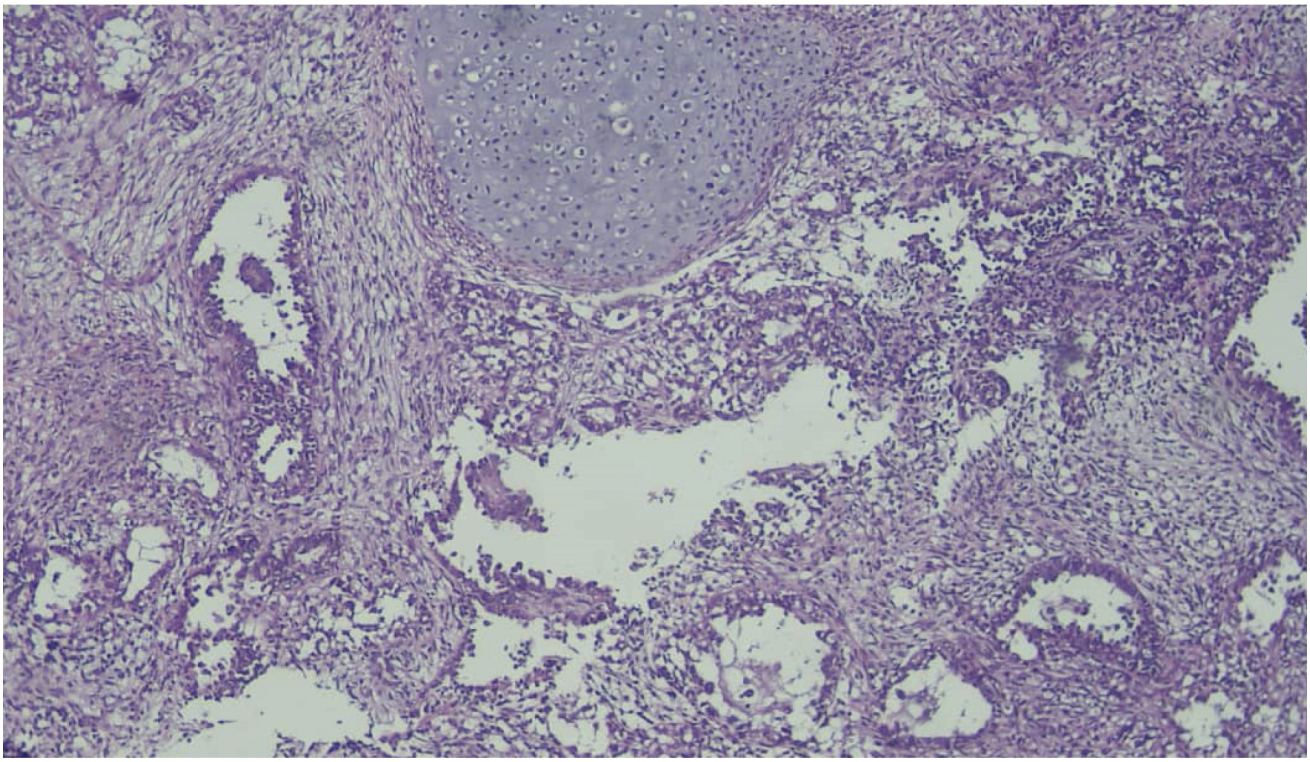

Figure 1. Histologic finding shows proliferation of atypical glands and sarcomatous component with cartilage formation (H & E staining, ×100).

margins and other tissues were tumor-free.

Ethical considerations

The patient signed an informed consent. The Ethical Committee of Mashhad University of Medical Sciences approved the present case report (ethics code: IR.MUMS.REC.1400.309).

Discussion

MMMT of the uterine cervix is rare. According to previous case reports, the median age of detecting this tumor is 65 years old. Almost all patients who refer to clinics have abnormal vaginal bleeding, abnormal Pap smear test result, or a cervical mass. The range of tumor size varies from 1cm to 10 cm.[1,4,5,6,7,8] The patient in this research was referred to our center due to unfavorable pathology findings in fractional dilatation and curettage plus postmenopausal bleeding, likewise other MMMT cases. MMMTs are biphasic malignancies that comprise an admixture of malignant epithelial and mesenchymal parts. The sarcomatous component may be homologous (fibroblast and smooth muscle) or heterologous (cartilage, striated muscle, or bone). Moreover, the epithelial part includes a variety of different histologic types, such as squamous cell carcinoma (SCC), adenocarcinoma, adenoid-squamous carcinoma, adenoid basal carcinoma, basaloid squamous carcinoma, adenoid cystic carcinoma, and undifferentiated carcinoma.[2,5,6,7] In this case, we observed carcinoid-carcinoma in the pathology report. A few articles are available which have reviewed former case reports of MMMTs in the uterine cervix. In 2012, Jong Joon Shim et al. studied a 45-year-old woman, with intermittent vaginal bleeding, a mass in the cervix, whose serum level of CA 125 was 5020 U/ml, based on paraclinical imaging (Computed tomography, MRI), which suggested the involvement of parameters, pelvic and para-aortic lymph nodes, and distant metastasis, such as hepatic nodules, diagnosed with uterine cervical cancer stage IV B. Later, a colposcopy punch biopsy, was performed and carcinoid sarcoma was established and MMMT was the final diagnosis.[2] Munakata et al., in 2013, reported another uterine cervix MMMT with neuroendocrine components.[4] A 43-year-old woman was referred to their center because of an abnormal Pap smear result. In cervical punch biopsy, SCC was defined. The patient then received adjuvant chemotherapy and radical hysterectomy type II was performed. Additionally, in the permanent pathology report, cervical SCC and rhabdomyosarcoma were mentioned. Therefore, chemotherapy continued for her, and in the follow-up after 38 months, they did not see any relapse.[4] Massinde et al., in 2012, published an article about MMMT in a prolapsed cervix.[3] Their case was a 37-year-old woman, with a history of seven vaginal deliveries, and two cervical polypectomies. She referred to their center with a bulging mass in her vagina. On the other hand, she suffered from stinky vaginal discharge and abnormal vaginal bleeding. In the pelvic examination, they found grade III prolapsed cervix and one round solid mass on it. Total vaginal hysterectomy was performed for her, and the pathology report demonstrated MMMT in the cervix. Vagina, adnexa, and lymph nodes were free of tumor.[3]

Conclusion

In summary, we described a case report of MMMT in the uterine cervix, with stage IBI. To the best of our knowledge, there are no specific symptoms for the diagnosis of MMMT, and early diagnosis is difficult. Almost all of them are diagnosed through pathological examinations and reports. Despite the fact that it is an aggressive kind of tumor, our case was successfully managed with rapid early diagnosis, surgery, and the combination of brachytherapy and chemoradiation therapy. However, further follow-up research is of great necessity.

Source of Funding

This study was supported by Mashhad University of Medical Sciences.

Data Availability Statement

The data that support the findings of this study

are available on request from the corresponding author.

Informed Consent

Written informed consent was obtained from the patient.

Conflict of Interest

None declared.

References

1. Zhang Q, Liu A, Wu JJ, Niu M, Zhao Y, Tian SF, et al. Primary malignant mixed Müllerian tumors of the fallopian tube with cervix metastasis: A rare case report and literature review. *Medicine* (*Baltimore*). 2018;97(28):e11311. doi: 10.1097/MD.0000000000011311.
2. Shim JJ, Jae CS, Hye KL, Kyoung EL, Ghi JL, Ho KK, et al. Malignant mixed Mullerian tumor arising from the uterine cervix: A case report. *J Korean Soc Radiol*. 2012;67(4):263-7. doi: 10.0000/jksr.2012.67.4.263.
3. Massinde AN, Rumanyika RR, Kihunrwa A, Rambau P, Magoma M. Malignant mixed Mullerian tumour of the prolapsed cervix: A case report. *Tanzan J Health Res.* 2012;14(2):158-61. doi: 10.4314/thrb.v14i2.10.
4. Munakata S, Iwai E, Tanaka T, Nakamura M, Kanda T. Malignant müllerian mixed tumor of the uterine cervix with a small cell neuroendocrine carcinoma component. *Case Rep Pathol.* 2013;2013:630859. doi: 10.1155/2013/630859.
5. Grayson W, Taylor LF, Cooper K. Carcinosarcoma of the uterine cervix: a report of eight cases with immunohistochemical analysis and evaluation of human papillomavirus status. *Am J Surg Pathol.* 2001;25(3):338-47. doi: 10.1097/00000478-200103000-00008.
6. Wright JD, Rosenblum K, Huettner PC, Mutch DG, Rader JS, Powell MA, et al. Cervical sarcomas: an analysis of incidence and outcome. *Gynecol Oncol*. 2005;99(2):348-51. doi: 10.1016/j.ygyno.2005.06.021.
7. Kudela E, Slavik P, Visnovsky J, Buocik P, Sivakova J, Sumichrastova P, et al. Malignant mixed Mullerian tumor of the cervix - Case report. *Cancer Treatment Communications.* 2014;2(1):12-5. doi: 10.1016/j.ctrc.2014.02.001.
8. Maheshwari A, Gupta S, Shet T, Wuntkal R, Tongaonkar HB. Diagnostic dilemma in a case of malignant mixed mullerian tumor of the cervix. *World J Surg Oncol.* 2006;4:36. doi: 10.1186/1477-7819-4-36.