



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Endometrial adenocarcinoma recurrence presenting with tibial metastasis: Report of a case



Mehmet Salih Söylemez^a, Bahattin Kemah^{b,*}, Umut Perçem Orhan Söylemez^c,
Bülent Kılıç^d, Korhan Ozkan^e

^a Bingol State Hospital, Department of Orthopaedics & Traumatology, Bingol, Turkey

^b Agri State Hospital, Department of Orthopaedics & Traumatology, Agri, Turkey

^c Bingol State Hospital, Department of Radiology, Bingol, Turkey

^d I. Gelişim University School of Health Sciences, Istanbul, Turkey

^e I. Medeniyet University Göztepe Training and Research Hospital, Orthopaedics and Traumatology Department, Istanbul, Turkey

ARTICLE INFO

Article history:

Received 14 January 2017

Received in revised form 16 April 2017

Accepted 16 April 2017

Available online 18 April 2017

Keywords:

Case report

Orthopaedic oncology

Bone metastasis

Solitary bone lesion

Endometrial adenocarcinoma

ABSTRACT

INTRODUCTION: Metastatic bone disease at extremities is mostly associated with lung, liver, prostat, thyroid or breast malignancies. There for surgeons generally tends to seek for a primary tumor originating from these organs. Herein a case of endometrial adenocarcinoma recurrence that presented with symptoms of tibial pain is described.

PRESENTATION OF CASE: 59 year-old woman was admitted to our orthopaedic oncology unit with pain, swelling and tenderness at right cruris for two weeks without any trauma history. Her medical history revealed that she had a total abdominal hysterectomy and bilateral salpingo-oophorectomy. During follow-ups no recurrence had been detected. Initial X-rays of the right tibia showed a lytic and expansile mass located at the shaft of the tibia suggesting metastasis. A wide resection of the lesion with clear margins was performed two weeks after first admittance. Resected area was replaced by fresh frozen femoral shaft allograft. At postoperative 17th month. X-rays obtained at last follow-up demonstrated full healing and integration of allograft.

DISCUSSION: Endometrial adenocarcinoma is a disease of postmenapausal women with 95% of the cases occurring after the age of 40 years. Patients with advanced or recurrent endometrial cancer often have distant metastases found within the lymph nodes, liver, and/or lung.

CONCLUSION: Recurrence of endometrial cancer as a solitary bone lesion is a rare situation. Wide resection and reconstruction with an allograft or an intercalar prosthesis might be an option to increase survival and possible cure of the patient.

© 2017 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Endometrial adenocarcinoma is one of the gynecological malignancies which tends to occur in postmenopausal women. Irregular uterine bleeding is the most common clinical presentation of these adenocarcinomas. Malignancies originating from uterine tissue can metastase to several organs with a predilection to lungs and liver. Metastasis to skeletal system is extremely rare and mostly have been reported to be in axial skeleton [1].

Metastatic bone disease at extremities is mostly associated with lung, liver, prostat, thyroid or breast malignancies. There for sur-

geons generally tends to seek for a primary tumor originating from these organs [1,2]. However although it is very rare, solitary bone lesions has been reported to be even first presentation site of a previously cured endometrial adenocarcinoma. Herein a case of endometrial adenocarcinoma recurrence that admitted to our university hospital with symptoms of tibial pain is described.

2. Presentation of case

59 year-old Turkish woman was admitted to our orthopaedic oncology unit with pain, swelling and tenderness at right cruris for two weeks without any trauma history. She had no any other complaint and she had walked into the clinic. She did not had a history of hereditary genetic disorder and she was not smoking. Her medical history revealed that she had a total abdominal hysterectomy and bilateral salpingo-oophorectomy with the diagnosis of endometrial adenocarcinoma followed by chemotherapy two years ago. During follow-ups no recurrence had been detected. Initial X-rays of the

* Corresponding author.

E-mail addresses: slhsylmz@gmail.com (M.S. Söylemez), bahattinkemah.md@gmail.com (B. Kemah), umutpercem@gmail.com (U.P.O. Söylemez), dr.buletk@hotmail.com (B. Kılıç), korhanozkan76@gmail.com (K. Ozkan).



Fig. 1. Preoperative anterior-posterior and lateral X-rays showing lytic expansile mass located at the shaft of the tibia.

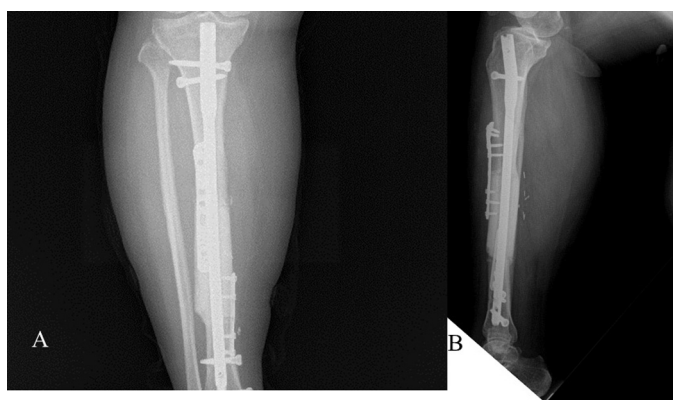


Fig. 2. Anterior-posterior and lateral X-rays showing tibia healed at postoperative first year.

right tibia showed a $3 \times 1,5$ cm lytic and expansile mass located at the shaft of the tibia suggesting metastasis (Fig. 1). MRI images demonstrated contrast enhanced $3 \times 1,5$ cm medullary expansile lesion surrounded by bone edema. A whole body PET/CT was performed to detect any other possible lesions. However PET/CT imaging didn't demonstrate any increased FDG uptake at any other location. A true-cut biopsy was undertaken which revealed an adenocarcinoma consistent with a primary endometrial tumor.

A wide resection of the lesion with clear margins was performed two weeks after first admittance. Resected area was replaced by fresh frozen femoral shaft allograft and was fixed with intramedullary nail and plate. Osteotomy lines was supported by impacting autograft obtained from iliac crest. The patient underwent additional adjuvant chemotherapy. At postoperative 3th. month weight bearing was allowed and no recurrence was detected at postoperative 17th month. X-rays obtained at last follow-up demonstrated full healing and integration of allograft (Fig. 2).

3. Discussion

Metastatic bone disease is the most common malignancy of bone. The most common primary cancer site is the lung (50%), followed by the colon, breast and genito-urinary tract [2]. Endometrial adenocarcinoma is a disease of postmenopausal women with 95% of the cases occurring after the age of 40 years [3]. Patients with advanced or recurrent endometrial cancer often have distant metastases found within the lymph nodes, liver, and/or lung [3,4]. However, there have been only few cases reported of pri-

mary endometrial cancer with metastasis to the bone. It has been reported to vary 2–6% [5]. In an autopsy study, the corresponding frequency was 25–75% [6]. Bone metastasis are mostly seen as recurrence and survival is better when compared to those who sustained solitary bone metastasis at primary diagnosis [1,7].

Acral metastases (metastases distal to the elbow and the knee) after endometrial adenocarcinoma is very rare. Bone metastases are mostly seen in vertebral column and pelvic ring because of batson venous plexus which drains periuterine and paravertebral region [3,5,6,8]. However, in rare cases metastasis to talus, calcaneus, tarsus and femur had also been reported [4]. Endometrial adenocarcinoma presenting with a bone metastasis at the first diagnosis is rarely seen. Usually first sign is abnormal uterine bleeding and pelvic pain, yet endometrial adenocarcinoma diagnosed after tibial metastases have been reported in 7 cases up to now [1,3,9–13]. Metastatic bone disease is usually seen after first diagnosis of endometrial adenocarcinoma and duration between diagnosis and metastasis is 6 months to 5 years [6]. However recurrence of the disease presented with isolated tibial metastasis after treatment is very rare and this is 5th. case that have been reported up to now in the english speaking literature [8,11,14].

Isolated bone metastasis after endometrial adenocarcinoma is seen in 50% of the patients that have a bone metastasis and these metastases are mostly with low grade disease as high grade disease tends to be multicentric [8]. Treatment of metastatic bone tumors depends on staging and tumor extent. Amputative surgery for acral metastases may be included, for cancers with poor response to radiation therapy and chemotherapy [15]. Painful metastases to the fingers of both hands and feet have been advised to be treated with amputation to increase life comfort during the terminal period and to ease patient's daily life activities [16,14]. However as it was seen in our case if disease have been presented with an isolated solitary bone metastasis a surgical procedure may be effective to obtain a longer life expectancy while protecting the limb and possible cure of the disease. Although survival following metastatic bone disease have been reported to be dismal, our patient is well after 17 months post operatively with no recurrence [2,10]. Wide resection that is tumoral tissue removed with a margin of normal tissue may be the primary goal in patients with isolated metastases of endometrial cancer to control the disease progression. Our patient was very satisfied with her operation as she had no restriction in her life and was cured during that follow-up time.

4. Conclusion

Recurrence of endometrial cancer as a solitary bone lesion is a rare situation. Although solitary bone lesions of extremities are mostly associated with lung, liver, prostat, thyroid or breast malignancies, endometrial **cancer** should be kept in mind for differential diagnosis as well. Wide resection and reconstruction with an allograft or an intercalar prosthesis might be an option to increase survival and possible cure of the patient as seen in our case.

Conflict of interest

The authors declare that they have no competing interests.

Source of funding

Any study sponsors had no involvement.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. As these

was a report of an interesting case and was not a trial or an observational research there was no need for an ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Authors contribution

Korhan Ozkan, Bülent Kiliç, and Umut Perçem Orhan Söylemez contributed to the conception and design of the study, carried out the literature research, manuscript preparation and manuscript review.

Mehmet Salih Soylemez and Bahattin Kemah, were involved with the case and writing of the manuscript, general management of the patient and revised the manuscript for important intellectual content.

All authors read and approved the final manuscript.

Registration of research studies

Name of the registry: Endometrial Adenocarcinoma Recurrence Presenting With Tibial Metastasis: Report of A Case.

Unique identifying number (UIN): researchregistry2387.

Guarantor

Assoc. Prof. Dr. Korhan Ozkan.
Mehmet Salih Söylemez, MD.

Acknowledgement

This manuscript has been prepared and reported in line with SCARE criteria [17]. All authors declare that they have no conflict of interest.

References

- [1] A. Kaya, A. Olmezoglu, C.S. Eren, U. Bayol, T. Altay, L. Karapinar, et al., Solitary bone metastasis in the tibia as a presenting sign of endometrial adenocarcinoma: a case report and the review of the literature, *Clin. Exp. Metastasis* 24 (2) (2007) 87–92.

- [2] C.J. Flynn, C. Danjoux, J. Wong, M. Christakis, J. Rubenstein, A. Yee, D. Yip, E. Chow, Two cases of acrometastasis to the hands and review of the literature, *Curr. Oncol.* 15 (5) (2008) 51–58.
- [3] V. Loizzi, G. Cormio, A. Cuccovillo, N. Fattizzi, L. Selvaggi, Two cases of endometrial cancer diagnosis associated with bone metastasis, *Gynecol. Obstet. Invest.* 61 (1) (2006) 49–52.
- [4] J. Albareda, M. Herrera, A. Lopez Salva, J. Garcia Donas, R. Gonzalez, Sacral metastasis in a patient with endometrial cancer: case report and review of the literature, *Gynecol. Oncol.* 111 (3) (2008) 583–588.
- [5] A. Shigemitsu, N. Furukawa, N. Koike, H. Kobayashi, Endometrial cancer diagnosed by the presence of bone metastasis and treated with zoledronic acid: a case report and review of the Literature, *Case Rep. Oncol.* 3 (3) (2010) 471–476.
- [6] F.W. Abdul-Karim, M. Kida, W.B. Wentz, J.R. Carter, K. Sorensen, M. Macfee, et al., Bone metastasis from gynecologic carcinomas: a clinicopathologic study, *Gynecol. Oncol.* 39 (2) (1990) 108–114.
- [7] S.M. Kehoe, O. Zivanovic, S.E. Ferguson, R.R. Barakat, R.A. Soslow, Clinicopathologic features of bone metastases and outcomes in patients with primary endometrial cancer, *Gynecol. Oncol.* 117 (2) (2010) 229–233.
- [8] S. Uccella, J.M. Morris, J.N. Bakkum-Gamez, G.L. Keeney, K.C. Podratz, A. Mariani, Bone metastases in endometrial cancer: report on 19 patients and review of the medical literature, *Gynecol. Oncol.* 130 (3) (2013) 474–482.
- [9] O. Onuba, Pathological fracture of right tibia, an unusual presentation of endometrial carcinoma: a case report, *Injury* 14 (6) (1983) 541–545.
- [10] M. Ishibashi, R. Fujiwaki, I. Nakayama, H. Miura, K. Sawada, Endometrial carcinosarcoma presenting as a tibial metastasis, *Int. J. Clin. Oncol.* 12 (4) (2007) 305–308, Epub 2007 Aug 20.
- [11] X. Le loet, B. Chevallier, C. Ducastelle, B. Pinel, J.M. Thomine, P. Deshayes, A case of single tibial metastasis disclosing cancer of endometrium, *Rev. Rheum. Mal. Osteoartic.* 1983 (50) (1983) 246–247.
- [12] E.E. Pakos, D.N. Gartzonikas, P.G. Tsekeris, T.A. Xenakis, Solitary tibial osteolytic lesion, *Case Rep. Med.* 2009 (2009) 352085, Epub 2009 Jun 16.
- [13] G. Artioli, M. Cassaro, L. Pedrini, L. Borgato, L. Corti, A. Cappetta, et al., Rare presentation of endometrial carcinoma with singular bone metastasis, *Eur. J. Cancer Care (Engl.)* 19 (5) (2010) 694–698.
- [14] G. Armentano, P.L. Bracco, R. Brizio, G. Perelli, Untreated endometrial adenocarcinoma: a case report, *Eur. J. Gynaecol. Oncol.* 18 (2) (1997) 144–145.
- [15] A.F. Mavrogenis, G. Mimidis, Z.T. Kokkalis, E.S. Karampi, I. Karampela, P.J. Papagelopoulos, A. Armaganidis, Acrometastases, *Eur. J. Orthop. Surg. Traumatol.* 24 (3) (2014) 279–283.
- [16] S. Soylemez, M. Demiroglu, M.A. Yayla, K. Ozkan, B. Alpan, H. Ozger, Lung metastasis mimicking fingertip infection, *Case Rep. Oncol. Med.* 2015 (2015) 708789, <http://dx.doi.org/10.1155/2015/708789>.
- [17] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. RORgill, the SCARE group, The SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (2016) 180–186.

Open Access

This article is published Open Access at sciedirect.com. It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.