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THYROID METASTASIS OF VAGINAL LEIOMYOSARCOMA: A CASE REPORT AND REVIEW OF THE LITERATURE

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Metastasis of vaginal leiomyosarcoma to the thyroid gland is an extremely rare event. Here, we report a case of thyroid metastasis of vaginal leiomyosarcoma. A 65-year-old female patient presented with a multinodular toxic goiter. 5 years earlier the patient has undergone a radical vaginal resection followed by chemotherapy due to high-grade vaginal leiomyosarcoma. Fine-needle aspiration cytology of the right thyroid lobe was suggestive of the benign thyroid lesion. As the diagnosis was not clear, following a multidisciplinary team discussion the decision was made to proceed with a total thyroidectomy. Pathologic assessment of the tumor confirmed thyroid metastasis of vaginal leiomyosarcoma.

Key Words: **metastatic thyroid carcinoma, vaginal leiomyosarcoma.**

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Vaginal leiomyosarcomas are extremely rare neoplasms. Out of all malignant vaginal neoplasms, leiomyosarcomas account for about 2% [1–5].The main symptoms of the disease might be associated with the tumor location and size. Since only about 140 vaginal leiomyosarcomas have been reported during the past 40 years, guidelines for the determination of disease stage and treatment have not yet been established [4]. At the same time, the thyroid gland is an uncommon site for metastatic disease. Here, we present a rare case of a patient with large primary vaginal leiomyosarcoma metastases to the thyroid and review the evidence on the role of thyroid surgery in the context of the current literature.

Case report. A 65-year old woman presented with vaginal bleeding and progressively enlarging mass in her left buttock that was first noted in 2007. Menopause had occurred when she was 49 years old. She had a history of 3 spontaneous deliveries and 2 abortions. Her past medical history was significant for hysterectomy performed because of uterine leiomyoma and removal of bilateral mammary fibroadenomas. She had no family history of cancer or other diseases. At the time of admission, her vital sign was stable and her general condition was good. Her hemoglobin concentration was 11.3 g/dL, blood cell volume was 32.4%, and platelet count was 331,000/ mm^3. On physical examination, the spherical mass close to her uterine rectal nest was detected and she was referred to the Department of General Surgery at our hospital.

On computed tomography, a mass sized approximately 9.2 × 8.4 cm was detected in her uterine rectal nest. There was no obvious pelvic lymph node enlargement and no pelvic effusion.

Our patient was prepared for surgery following our routine preoperative procedures. Placed in the dorsall lithotomy position she underwent the resection of vaginal tumor. The tumor, including the capsule, was resected completely; and a drain was inserted into the area from which the tumor had been removed. A complete macroscopic excision of tumor sized 9 cm was achieved.

Based on the histopathological results, the tumor was diagnosed as a high grade leiomyosarcoma. The tumor cells comprised spindle-cell nuclei with blunt ends, coarse nuclear chromatin, and prominent nucleoli. Surgery was followed by adjuvant chemotherapy consisting of gemcitabine/docetaxel x 4 cycles. She then subsequently relapsed with metastatic disease, thyroid nodules were noted on ultrasonography of the neck 5 years later. She presented with hypertension, high heart rate, neck swelling with intermittent neck discomfort without airway pressure symptoms. On clinical examination she was found to have multinodular toxic goiter. Laboratory findings revealed thyroid-stimulating hormone — < 0.005 IU/mL. The neck swelling was investigated with an ultrasound and confirmed multinodular toxic goiter. The fine-needle aspiration cytology (FNAC) of the right thyroid lobe was described as more suggestive of the benign thyroid lesion. As the diagnosis was not clear, following a multidisciplinary team discussion the decision was made to proceed with a total thyroidectomy. Histopathological analysis of the specimen revealed metastasis of vaginal leiomyosarcoma (Fig. 1, 2). The overall appearances were consistent with metastatic vaginal leiomyosarcoma with no evidence of a primary thyroid malignancy. Chemotherapy was performed according to the following regimen: gemcitabine 1000 mg/m^2 and docetaxel 60 mg/m^2, for a total of 4 cycles. Our patient was clinically well 15 months following thyroidectomy with follow-up computed tomography imaging showing no evidence of local

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Abbreviation used: FNAC — fine-needle aspiration cytology.

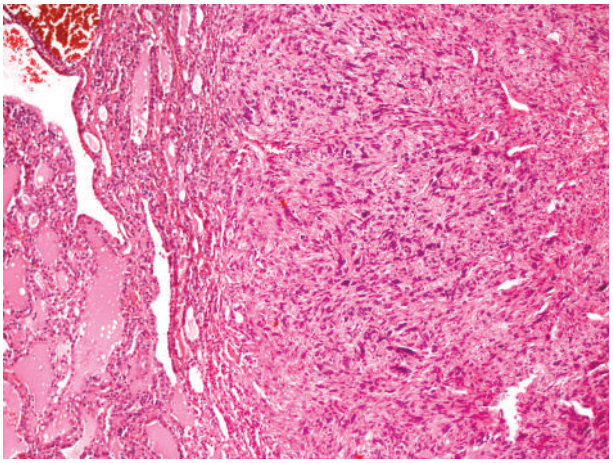

Fig. 1. Histopathology section showing metastatic thyroid carcinoma, metastasis of vaginal leiomyosarcoma. Hematoxylin-eosin. × 200

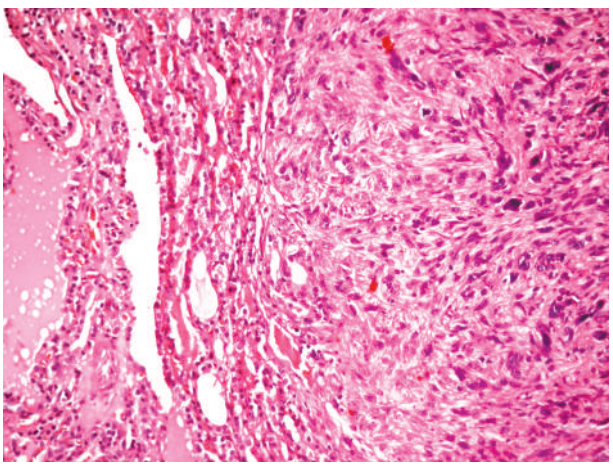

Fig. 2. Histopathology section showing metastatic thyroid carcinoma, metastasis of vaginal leiomyosarcoma. Hematoxylin-eosin. × 400

recurrence. After that the patient's general condition deteriorated rapidly. 15 months after thyroidectomy the patient died because of multiorganic failure.

Review of the literature. Smooth muscle is a component of many tissues and organs. So, leiomyosarcoma can arise at almost any organ [2]. The average age at diagnosis is around 50 years [2]. Preoperative diagnosis is challenging, and tumors are usually diagnosed by histopathology after resection [6]. The most frequent histological variants of vaginal tumors are squamous cell carcinomas (up to 90%), adenocarcinomas (up to 10%), melanomas (3%), and sarcomas (3%). Malignancy of leiomyosarcomas is diagnosed histopathologically by assessment of cellular mitotic activity [4]. Nuclear atypia, mitotic index, and zonal necrosis are parameters used in the differentiation from leiomyomas [7]. It is generally accepted that the presence of 10 or more mitoses per 10 high power fields indicates malignancy [7]. Due to its rarity, vaginal leiomyosarcoma is seldom detected prior to surgery, and typical symptoms on physical examination have not been described. The major clinical manifestations are a growing vaginal mass, abdominal pain and vaginal bleeding.

Vaginal smooth muscle tumors mostly develop in the anterior vaginal wall [2]. In this case, the tumor developed in her posterior vaginal wall, a location with high predilection for malignant neoplasms. Differential diagnosis of these vaginal masses include Gartner's cyst, granuloma, epithelial carcinoma, adenocarcinoma, rhabdomyosarcoma, melanoma, small cell carcinoma and Müllerian sarcoma that can develop in the vagina [4]. Primary treatment typically consists of resection of the tumor with adequate margins [3].

After resection of the tumor, the ideal management is less well defined. While surgery is considered the mainstay of treatment, there was no difference in survival of patients who had surgery alone and those who had surgery followed by adjuvant radiotherapy or chemotherapy [8]. Due to its extremely aggressive behavior and a great tendency for local recurrence and distant metastasis, the 2-year survival rate is less than 50% [9, 10]. The prognostic factors include age, tumor size, histological grade, International Federation of Gynecology and Obstetrics stage, mitotic count, tumor injury, and treatment modality. Tumors usually relapse within 2 years with distant metastases to lung, breast or liver [5, 11].

No studies have evaluated vaginal leiomyosarcoma accompanied with thyroid metastasis. Metastatic deposits have a predilection for highly vascularized organs but despite one of the highest blood supplies the thyroid is rarely the site of metastatic deposits [6, 12]. The exact mechanism for the lack of metastasis to the thyroid is unknown, but it is thought to be associated with immunological mechanisms [13].

Diagnosis of thyroid metastasis is challenging, and most cases are asymptomatic and identified due to the appearance of solitary or multiple masses [13]. Recently, diagnoses of metastatic thyroid glands have increased due to development of diagnostic techniques such as ultrasonography and FNAC. However, in the reported case FNAC didn't help with the diagnosis of metastasis of thyroid gland.

Metastases to the thyroid gland represent an indication for surgery in < 1per 1000 thyroidectomies [14, 15] of which around 50% are from a primary renal cell carcinoma [16]. The rest of primary neoplasms that have been reported to metastasize to the thyroid include colorectal, lung, breast, malignant melanoma and gastrointestinal tumors [17–20]. Thyroid metastasis of a primary cancer is usually considered as a systematic disseminated disease with a generally poor prognosis [13, 21, 22]. Survival following a diagnosis of thyroid metastasis varies depending on the site of the prior malignancy [13].

Surgical methods are rarely mentioned in the literature, to the best of our knowledge there are no prospective studies dedicated to the role of surgery in metastatic disease of the thyroid. In the present case report the patient with vaginal leiomyosarcoma metastasis to the thyroid underwent total thyroidectomy with good outcome. According to the previous studies results, isolated thyroidectomy was the operation of choice as a local disease control option to palliate

and prevent the potential morbidity of tumor extension to the airway [23].

CONSENT FOR PUBLICATION

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review.

COMPETING INTERESTS

The authors declare that they have no competing interest.

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