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Author(s)	SHIMIZU, Yousuke; OGAWA, Osamu; TERACHI, Toshiro; OKADA, Yusaku; YOSHIDA, Osamu
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A CASE OF PRIMARY URETHRAL LYMPHOMA PRESENTING AS A HUGE MASS SURROUNDING THE FEMALE URETHRA

Yousuke SHIMIZU, Osamu OGAWA, Toshiro TERACHI,
Yusaku OKADA and Osamu YOSHIDA

From the Department of Urology, Faculty of Medicine, Kyoto University

A patient with a primary malignant lymphoma surrounding the female urethra is reported. Despite the good response of the primary tumor to radiotherapy, the patient died shortly after diagnosis due to disseminated disease. We reviewed 16 cases of this rare entity reported previously.

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Key words : Female urethra, Malignant lymphoma, Radiotherapy

INTRODUCTION

Malignant lymphoma can affect almost any organ of the body, although it usually originates in the lymph nodes. The incidence of malignant lymphomas in urogenital organs is low. Limited clinical experience has resulted in uncertainty concerning the most appropriate form of therapy for this malignancy. We report an 82-year-old woman with non-Hodgkin's lymphoma presenting as a huge tumor surrounding the female urethra.

CASE REPORT

An 82-year-old woman consulted our hospital with complaints of dysuria and sense of residual urine

which had persisted for 2 months. Physical examination revealed an elastic hard mass along the urethra. Laboratory examination revealed no abnormal findings except the elevation of serum level of lactic dehydrogenase (555 IU/L; normal < 230). Cystoscopy revealed the bladder neck elevated by the mass around the urethra, although the bladder mucosa was smooth, indicating that the tumor had originated from the paraurethral region. Gynecological examination did not reveal any abnormal findings in female genital organs. Pelvic computed tomography (CT) and magnetic resonance imaging (MRI) revealed a 5.5 cm × 5 cm × 5.2 cm mass surrounding the urethra (Fig. 1). Cold cup biopsy was performed, and the histopathological examination showed infiltration of poorly differentiated malignant cells (Fig. 2). After immunohistochemical staining of the specimen (CD20 (+), CD45RO (-), CD3 (-), CD15 (-), CD68 (-)), the tumor was diagnosed as non-Hodgkin's lymphoma, B cell type, diffuse and large cell type according to the Lymphoma Study Group in Japan (LSG) classification¹⁾. There was no evidence of tumor elsewhere under radiological examination. The



Fig. 1. MRI revealed a large homogeneous mass (5.5 cm × 5 cm × 5.2 cm) surrounding the urethra.

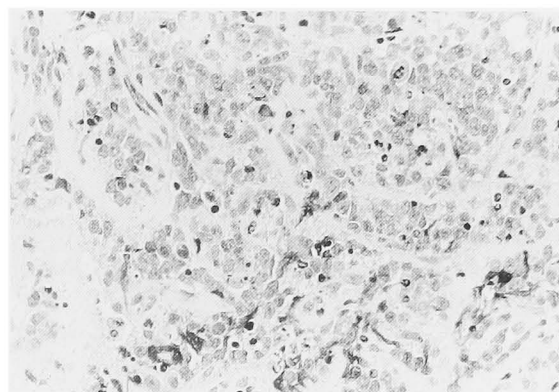


Fig. 2. Histological examination of the biopsy specimen revealed proliferation of small and large malignant cells that were diagnosed as lymphoma cells by immunohistochemical analysis.

bone marrow aspiration biopsy failed to show the infiltration of lymphoma cells. Under the diagnosis of locally invasive urethral malignant lymphoma, the patient underwent 40 Gy radiotherapy to the whole pelvis and the bilateral inguinal region. Although the primary lesion responded well to the radio-



Fig. 3. MRI after radiotherapy revealed a remarkable response in the primary tumor.

therapy (Fig. 3), multiple lung metastases appeared during the therapy. The patient died of disseminated disease five months after radiotherapy despite oral administration of 25 mg/day of etoposide.

DISCUSSION

According to Freeman's study²⁾, primary urogenital malignant lymphomas are relatively rare, and comprised 2.6% of all extranodal lymphomas. To the best of our knowledge, only 16 cases of urethral lymphoma, including 2 male patients, have been reported, all being histopathologically diagnosed as non-Hodgkin lymphomas (Table 1). Urethral lymphomas usually present with a painless mass resembling a caruncle. This patient, however, had a huge mass beneath the bladder neck indicating infiltration to the periurethral tissue. This particular pattern of tumor growth was the cause of her major complaint of dysuria with a sense of residual urine.

There is no consensus about treatment of urethral lymphoma. Various treatments have been applied to urethral lymphoma. Of the 16 patients reported previously 12 had no evidence of disease at 31.7 months of mean follow-up. Although the number of patients was very small and the follow-up period was short in all cases, local therapy including excision or external beam radiation with/without chemotherapy showed a good outcome in patients with local or locally invasive tumor. However, patients with disseminated disease showed a poor prognosis regardless of the kind of initial treatment. The

Table 1. Summary of reported cases of primary urethral lymphoma

Age	Presentation	Stage	Treatment	Followup	Reference
?	Unknown	Unknown	Radiation	NED, 9 mos.	[3]
83	Dysuria, Meatal mass	Local	None	Dead of unrelated cause	[4]
51	Urethral polyp	Local	Excision, Chemotherapy	NED, 10 yrs.	[5]
76	Large urethral mass	Local	Excision	NED, 2 yrs.	[6]
31	Spotting, Firm urethra	Local	Radiation, Chemotherapy	NED, 9 mos.	[7]
63	Urethral mass	Local	Excision, Chemotherapy	NED, 3 yrs.	[8]
78	Polyuria, Dysuria	Local	Chemotherapy	Unknown	[9]
65	Vaginal spotting	Local	Excision	NED, 2 yrs.	[10]
76	Caruncle, Hematuria	Locally invasive	TUR, Radiation	NED, 1 yr.	[11]
63	Vulvar pruritus, Dysuria	Locally invasive	Chemotherapy	NED, 4 yrs.	[12]
75	Vaginal spotting	Locally invasive	Radiation	NED, 42 mos.	[13]
62	Caruncle, Spotting	Disseminated	Excision	Dead, 5 mos.	[14]
53	Caruncle	Disseminated	Palliative	Dead, 2 mos.	[15]
77	Vaginal mass, Hematuria	Disseminated	Chemotherapy	NED, 9 mos.	[16]
57	Hematuria, Difficult voiding	Disseminated	Chemotherapy	Dead, 3 mos.	[17]
57	Urinary retention	Disseminated	Chemotherapy	NED, 6 mos.	[18]
82	Dysuria	Locally invasive	Radiation, Chemotherapy	Dead, 7 mos.	Present case

[16] and [17] are males. NED=No evidence of disease

present patient died only seven months after diagnosis despite a good response of the primary tumor to external radiotherapy. According to the Rappaport grading system or the Working Formulation, diffuse histology and high grade tumor are considered to show a more unfavorable outcome than nodular subtype and intermediate grade tumors. In patients with these types of unfavorable histology or disseminated disease, early intensive chemotherapy is recommended.

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和文抄録

尿道原発と考えられた悪性リンパ腫の1例

京都大学医学部泌尿器科学教室（主任：吉田 修教授）

清水 洋祐, 小川 修, 寺地 敏郎

岡田 裕作, 吉田 修

82歳女性。主訴は排尿困難と残尿感。膀胱底部から尿道周囲にかけて鶏卵大の腫瘤を認め、生検にて非ホジキンリンパ腫と診断された。他臓器に病変を認めなかったため尿道原発の悪性リンパ腫と診断し放射線療法を施行した。治療後、腫瘍は著明に縮小したが肺転移が出現した。エトポシド単剤による化学療法を追加したが、放射線治療後5カ月で腫瘍の急激な増加を認

め癌死した。本症例は国内外の文献上17例目の尿道原発悪性リンパ腫と考えられた。17例の治療成績をみると、限局性および局所浸潤例は切除、放射線、化学療法いずれも有効であったが、播種性のものは治療法に関わらず予後不良であった。

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