Hodgkin’s lymphoma of the surrenal: case report and review of the literature

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ABSTRACT

Lymphomas are the rare tumors of the surrenal gland. Among the lymphomas, non-Hodgkin’s lymphomas are seen more frequently while Hodgkin’s lymphoma is very rare. Here we reported a case with Hodgkin’s lymphoma involving surrenal gland and we reviewed the literature.

INTRODUCTION

Surrenal lesions are commonly determined as an incidental finding during computerized tomography (CT) and magnetic resonance imaging (MRI) in different issues. Most of these lesions are benign adrenal cortical adenomas. The surrenal gland is a frequent site of metastatic disease [1]. Among the solid tumors, lung, renal, breast and gastrointestinal cancers are the most frequent tumors metastasizing to the surrenal gland. Lymphomatous involvement of the surrenal is seen less commonly [2]. Lymphomas of the surrenal glands are generally seen in advanced stage disease and most of the reported cases are non-Hodgkin’s lymphomas [3]. Non-Hodgkin’s lymphoma occurs in surrenal in nearly 25% of patients during the course of disease. In contrast, Hodgkin’s lymphoma of the surrenal gland is very rare. Except autopsy series, so far only three cases have been reported [1, 4, 5]. Here a case with Hodgkin’s lymphoma involving surrenal gland has been reported and literature has been reviewed.

MATERIAL AND METHODS

Rigorous electronic literature searches were conducted to identify reports of surrenal Hodgkin’s lymphoma. Articles were identified in electronic database searches of Pubmed, online Turkish database, using a predetermined search strategy including the following terms: surrenal hodgkin’s lymphoma, adrenal gland and Hodgkin’s lymphoma. Broad search terms ensured that no cases were excluded inadvertently. At this search we found total of three articles published between 1950 and 2010.
CASE REPORT

39 year-old man admitted to the hospital with fatigue. Abdominal ultrasonography and CT showed a solid lesion with 56x58 mm diameter at the right surrenal region. Patient was operated, right nephrectomy and surrenalectomy were performed. Histopathological examination of surrenal mass was reported as Classical Hodgkin’s lymphoma-Nodular sclerosing type. CD30 was found to be positive in Reed Sternberg cells. CD20, CD3, LCA, S100, ALK, and chromogranin were found to be negative. EBER ISH was found to be positive. PET CT staging showed generalized lymph nodes located at mediastinum, axillary, paravertebral, infraabdominal and iliac regions. Bone marrow aspiration and biopsy were negative for Hodgkin’s lymphoma infiltration. Patient was treated by ABVD combination chemotherapy including doxorubicin, bleomycin, vinblastine and dacarbazine. PET CT after six cycles showed complete remission.

DISCUSSION

Surrenal gland is a frequent site for various tumors including benign and malignant types. The majority of these tumors are non-functional benign tumors. Approximately 1 in 4000 surrenal tumors has been found to be malignant [6]. Metastasis to the adrenal glands is a frequent finding at autopsy and most commonly occurs in patients with lung, gastrointestinal and renal cell cancer [7].

Lymphoma infiltration in surrenal gland is generally seen at the advanced stage of the lymphomas and is reported in 25% of the cases. Surrenal involvement is generally seen in non-Hodgkin lymphomas and may be seen as primary surrenal lymphomas or may accompany to systemic lymphomas. Primary surrenal lymphoma describes the term lymphoma limited to the surrenal gland without lymph node involvement and leukemic table. Primary surrenal lymphoma is the 3% of all extranodal lymphomas [3, 8]. In a retrospective analysis involving 435 autopsies and 29 surrenalectomy materials, histopathological examination showed 31 lymphomas (6.7%) and only 3 of these (0.65%) were Hodgkin’s lymphoma [9]. In another autopsy study surrenal involvement by Hodgkin’s lymphoma has been found to be 8% of the cases but information about this matter is limited [10, 11].

Table 1: Surrenal Hodgkin’s lymphoma cases (literature review)

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>Age</th>
<th>Gender</th>
<th>Size (cm)</th>
<th>Adrenal Insufficiency</th>
<th>Stage</th>
<th>Unilateral or Bilateral</th>
<th>Lymph Node involvement</th>
<th>Bone Marrow involvement</th>
<th>Patologic Subtype</th>
<th>CD15</th>
<th>CD20</th>
<th>CD30</th>
<th>Epstein Barr Virus</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Balci (1)</td>
<td>2009</td>
<td>69</td>
<td>M</td>
<td>9x6.5x2.5</td>
<td>No</td>
<td>III</td>
<td>U</td>
<td>Yes</td>
<td>No</td>
<td>Nodular Sclerosis</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>?</td>
<td>Adrenalectomy</td>
</tr>
<tr>
<td>Andrew (2)</td>
<td>2009</td>
<td>76</td>
<td>F</td>
<td>5.5x4.5x3.0</td>
<td>No</td>
<td>III</td>
<td>U</td>
<td>Yes</td>
<td>No</td>
<td>Nodular Sclerosis</td>
<td>+</td>
<td>+</td>
<td>(Variable)</td>
<td>?</td>
<td>Adrenalectomy</td>
</tr>
<tr>
<td>Ouansafi (3)</td>
<td>2010</td>
<td>76</td>
<td>F</td>
<td>4.7x2.3</td>
<td>?</td>
<td>III</td>
<td>U</td>
<td>Yes</td>
<td>No</td>
<td>Mixed cellularity</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>Right Adrenalectomy</td>
</tr>
<tr>
<td>Our Case</td>
<td>2011</td>
<td>39</td>
<td>M</td>
<td>6x5x4</td>
<td>No</td>
<td>III</td>
<td>U</td>
<td>Yes</td>
<td>No</td>
<td>Nodular Sclerosis</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>Right surrenal nefrectomy, Adjuvan Chemoterapy</td>
</tr>
</tbody>
</table>

When we exclude the autopsy series, surrenal involvement in Hodgkin’s lymphoma is very rare and so far only three cases have been reported in English literature (Table 1). We are reporting the forth case about this. Knowledge about the clinical outcome of cases with surrenal
involvement by Hodgkin’s lymphoma is very limited due to the very low number of these cases. Two of these 3 cases were female and their ages were 76, 76 and 69. Our case was very young (39 years old) as compared with reported cases. Symptoms of the cases were fatigue, weight loss and back pain. Surrenal insufficiency has not been reported in two cases, as seen in our case. Lymph node involvement has been reported in two of the reported cases and also in our case. It is very well known that metastatic surrenal tumors are bilateral in 75% of the cases. All the surrenal Hodgkin’s lymphoma cases reported and ours were unilateral and right sided. Bone marrow involvement has not been found in none of them. Pathologically 3 cases, including ours, had nodular sclerosing type and one case had mixed cellularity type. CD20 was found to be positive in 2 of four cases. CD30 was positive in all of the four cases. EBV was positive in three of the cases and this is an important finding.

Treatment for the surrenal Hodgkin’s lymphoma is not clear due to the very low frequency of this disease. Surgery has been performed in all of these cases, including ours, this was the necessity to diagnose this surrenal mass. Systemic chemotherapy is necessary to these cases as all other cases with extranodal Hodgkin’s lymphoma. The role of local radiation treatment is not known.

In conclusion we have limited information about the etiopathogenesis, prognosis and clinical outcome of surrenal Hodgkin’s lymphoma. Suitable treatment protocols can be proven with participate of wide clinic series. Furthermore, when diagnose this disease with biopsy systemic chemotherapy will be the preferred strategy instead of surgery as the main treatment. Also we can say that Hodgkin’s lymphoma must be considered in the differential diagnosis of surrenal masses.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

REFERENCES