Intraparenchymal chordoid Meningioma After Radiotherapy for Hodgkin Lymphoma: A Case Report and Review of the Literature

Mustafa Efendioglu¹, Recep Basaran¹, Dogan Gundogan², Fatih Han Bolukbasi³, Mustafa Kaksi⁴, Aydin Sav⁵ and Tuncay Kaner²

¹Dr. Lutfi Kirdar Kartal Training and Research Hospital, Department of Neurosurgery, Istanbul, Turkey
²Istanbul Medeniyet University Goztepe Education and Research Hospital, Department of Neurosurgery, Istanbul, Turkey
³Medipol University School of Medicine, Department of Neurosurgery, Istanbul, Turkey
⁴Yalova Hospital, Department of Neurosurgery, Yalova, Turkey
⁵Acibadem University School of Medicine Department of Pathology, Istanbul, Turkey

Abstract: Objective: Hodgkin lymphoma can be treated by radiotherapy or chemotherapy alone or combined. Meningiomas account for 1-4.2% of all primary intracranial tumors in children, and chordoid meningioma is a very rare subtype. In this study, we investigated a case of an intraparenchymal chordoid meningioma that developed during the early stage in a patient with Hodgkin lymphoma who had been treated with radiotherapy.

Case: A 10-year-old male patient was diagnosed with Hodgkin lymphoma and was treated with a combination of radiotherapy and chemotherapy. He presented at our emergency service 6 years later. He had a fever and was suffering from discomfort and insignificant left hemiparesis (4/5). Contrast-enhanced cranial magnetic resonance imaging (MRI) showed a mass in the right temporoparietal region. The intracranial lesion was surgically excised. The tumor was identified as a WHO grade 2 chordoid meningioma by the pathological examination. The Ki-67 proliferation index was found to be 20-25%.

Conclusion: Surgeons must remember that radiation-associated meningiomas may occur in the early stage of the treatment as well as in the late stage. Young patients with grade 2 chordoid meningiomas must be followed-up in case of recurrence, and tumors with high Ki-67 indexes are highly expected to relapse.

Keywords: Meningioma, Hodgkin lymphoma, radiation, secondary tumor, chordoid, intraparenchymal.

INTRODUCTION

Hodgkin lymphoma (HL) accounts for 1% of all carcinomas and 25-40% of lymphomas [1-5]. HL is common in males at all ages and peaks between the ages of 20 and 40 years [3, 6].

HL is classified by WHO grading [7]. The most common type in Turkey and developing countries is the mixed cellular type of HL [8].

Both chemotherapy (KT) and radiotherapy (RT) are effective methods for the treatment of Hodgkin lymphoma. Therefore, RT and KT alone or in combination can be applied [9-12].

Many of the complications that occur during the late stage after the treatment of Hodgkin lymphoma are associated with radiotherapy. The annual rate of secondary malignancy is approximately 1% [13, 14]. Despite this low rate, the secondary malignancies are the primary cause of mortality and morbidity in patients with HL [15, 16].

Meningioma is very rare in children. It accounts for 1-4.2% of all primary intracranial tumors during childhood [17-28]. Intraparenchymal or subcortical meningiomas without dural connections are even rarer [29].

Chordoid meningiomas accounts for only 0.5-1% of all meningiomas [30]. As with all meningiomas, chordoid meningiomas are generally more common in adults and are very rare in children [26, 31]. The development of a radiation-associated meningioma is rare and occurs long after radiation treatment [32, 33]. A radiation-associated cerebral meningioma, which is a very rare complication of leukemia treatment, has been reported [19, 20, 29, 34, 35]; however, only one case of cerebral meningioma after Hodgkin lymphoma treatment has been reported [36]. Posterior fossa meningioma, which is a type of meningothelial tumor, was also found.

In this article, we present the case of a patient who received radiotherapy for the treatment of Hodgkin...
lymphoma and was later diagnosed with a supratentorial meningioma during the early stage after treatment.

CASE REPORT

A 10-year-old male patient was examined in 2006 because of a nodule in his neck. He was diagnosed with mixed-cell, stage 4A classic Hodgkin lymphoma. The patient was treated with a combination of radiotherapy at 180 cGy/fr 25 Gy and 6 courses of ABVD chemotherapy in his bilateral neck and mediastinum. He was diagnosed with anemia prior to chemotherapy, and his hemoglobin level increased after treatment. The follow-up of the patient was performed for two years but was then terminated. The patient had no complaints within that period and then presented at the emergency service 6 years after the termination of radiotherapy. He had fever, impaired general condition and weakness on the left side. No pathological findings were detected in the physical examination except for insignificant left hemiparesis (4/5). The blood tests revealed only anemia. The patient had a resistant fever. PET and CT scans were repeated, and a slightly hypermetabolic mass with intense edema was observed in the right parietal space. In addition, a decrease in the metabolism of the right cerebral cortex was observed compared with its left-side counterpart. Contrast-enhanced cranial magnetic resonance imaging (MRI) revealed a mass with maximum dimensions of 49x41x39 mm located in the right temporoparietal region with significant pathological contrasting in the solid component (Figure 1). The doctors held a consultation and planned the surgical treatment.

SURGERY

A craniotomy was performed in the right temporoparietal region. The location of the mass was determined with sonography. We moved transcortically with bipolar coagulation and aspiration and reached a highly solid deeply located tumoral mass with cleavages in some areas. The tumoral tissue was tattletale gray, highly vascular and not absorbable due to the numerous feeding arteries. The solid tumoral mass was totally excised with CUSA (cavitron ultrasonic surgical aspirator).

HISTOPATHOLOGY

Microscopic examination showed tumor consisting of tumor cells forming sheets in which the neoplastic cells demonstrate classic cytological features of meningothelium, i.e., round to oval nuclei, delicate chromatin, small solitary nucleoli, and frequent nuclear-cytoplasmic invaginations and/or homogenization. In patchy areas of tumor, the tumor cells are largely uniform; with oval nuclei with delicate chromatin that on occasion show central clearing, or the formulation of cytoplasmic-nuclear inclusions mimicking arachnoidal cap cells. The invaginations, also termed pseudoinclusions, appear as round, circumscribed, and intranuclear areas surrounded by marginalized chromatin. Another eye-catching feature was cords or trabeculae formed by eosinophilic, often vacuolated cells entrapped in an abundant extracellular mucoid.

Figure 1: Preoperative MRI scan with contrast enhancement reveals a mass located on right temporoparietal area within deep white matter. It has pathological contrast enhancement on its solid component and centrally contains a cystic necrotic area. Prominent vasogenic oedema is found around the mass.
matrix background. Although classic features of meningioma, such as whorls and psammoma bodies, are scant, if present at all, in examples with advanced chordoid features. There was an associated myxoid matrix forming pools and interspersing tumor cells giving rise to impression of chordoid features to the meningeal tumor (Figure 2).

By using conventional histochemistry staining procedure, AS-AB identified acid mucopolysaccharides in the matrix consisting myxoid features (Figure 3).

Immunochemistry revealed a variable immuno-reactivity of tumors cells, i.e., membranous EMA (Figure 4), focal areas of cytoplasmic and nuclear S-100, focal nuclear progesterone reactivity. No CD34 and pancytokeratin immunoexpressivity was seen. Ki-67 (MIB-1) index was 10% (Figure 5) and no p53 reactivity at all.

Therefore differential diagnosis including tumors with associated with abundant myxoid features mimicking chordoid features including schwannoma with myxomatous change, chordoma, cartilaginous tumor, and pleomorphic adenoma of salivary gland. Schwannomas typically have reticulin stain around individual cells and are strongly positive for S-100 protein. Chordomas originate from midline structures and are composed of pathognomonicphysaliphorous cells but some cells with more solid and eosinophilic cytoplasm are often present. The cells are embedded in an abundant, basophilic, metachromatic myxomatous matrix. They are strongly immunoreactive for
S-100 protein, cytokeratin, and also EMA. Chondrosarcomas show a mixed hyaline and myxoid type. The histologic grade is always low. Immunohistochemistry allows separation of chordoid meningioma from chondrosarcomas. Over 95% of cranial base chondrosarcoma are positive for S-100 protein but only a few of them are immunoreactive for EMA. They are negative for cytokeratin. Pleomorphic adenoma of salivary gland with abundant myxoid matrix has S-100 and EMA negativity whereas reacting with cytokeratin. By conclusion, immunohistochemical panel of tumor tissue was consistent with chordoid meningioma.

**FOLLOW-UP**

During the early post-op period, the patient's left hemiparesis improved, and the fever subsided. A neurological examination was performed 6 months later, and the results were considered normal. No recurrence was detected in the control cranial MRI (Figure 6). The blood test results were normal.

**DISCUSSION**

Hodgkin lymphoma is a malignancy of the lymphoid tissue that was defined by Sir Thomas Hodgkin in 1832 and is characterized by malignant Reed Stenberg and Hodgkin cells and characteristic cellular structures [1-4]. The purpose of the treatment is to cure patients with minimum complications. KT and RT alone or in combination are effective methods to treat the disease [9-12]. A large portion of the body is exposed to radiation with either RT alone or with RT in combination with KT. Many of the complications that occur during the late state of treatment are associated with radiation exposure [13, 14]. Although the disease can be controlled in over 70% of patients with the combined treatment approach [37], the risks of second primary tumors, cardiovascular disease, cerebrovascular disease and infection are increased [38-50]. Therefore, treatment with KT alone is preferred [51].

To consider a secondary neoplasia to be associated with therapeutic radiation, 1- the radiation must have been delivered in the tumoral area, 2- there must be a symptomless period between the radiation therapy and onset of symptoms due to the secondary tumor, 3- the diagnosis of the tumor must be histologically verified, and 4- the new lesion must be histologically differentiated from the primary lesion [52, 53].

Meningiomas and gliomas are the most common tumors in later years in the patients who have received therapeutic radiation. Gliomas are generally observed in the first 5 years, and the frequency of meningiomas increases with time [32, 33]. The first case of radiation-associated meningioma was reported by Mann et al. in 1953 [54]. The period between the radiation exposure and the development of the meningioma is approximately 9.5 years in children [55]. In the study performed by Primoz et al., the risk of secondary intracranial meningioma in the first 10 years was 0.53%, with risks of 1.20% in 20 years and 8.18% in 25 years [55]. Secondary tumors of the central nervous system were observed in 17% of patients treated with radiation therapy.
system (CNS) in patients who received radiotherapy for Hodgkin lymphoma and have been reported in the literature are listed in Table 1.

A large number of cases of meningioma after radiotherapy for leukemia have been reported in the literature [63]. However, the development of meningioma after treatment for Hodgkin lymphoma has been reported by only Deutsch et al. in 1999 [36]. The case involved a male patient who was histologically diagnosed with a posterior fossa meningothelial meningioma 25 years after being treated with therapeutic radiotherapy at 15 years old.

In our case, the patient was diagnosed with Hodgkin lymphoma in 2006 and received radiotherapy in the same year. He had a symptomless period of approximately 6 years and then developed an intraparenchymal intracranial meningioma in the right temporoparietal region. The intracranial lesion was surgically excised. Histological evaluation revealed that it was a chordoid meningioma. In contrast to cases of meningioma reported in the literature, our patient’s meningioma developed in a very short period of time after the radiation treatment. In addition, our patient’s tumor was a chordoid meningioma, which is a rare histological type. According to the definition of radiation-associated tumors, secondary tumors are observed in the areas exposed to radiation [53, 54]. In the study performed by Deutsch et al., the patient received radiotherapy in the supine position and developed a tumor in the posterior fossa due to excessive radiation exposure because the radiation beams were aimed directly at the posterior fossa, resulting in the accumulation of direct and reflected beams (Figure 4) [36].

A review by Primoz et al. revealed that the development of second primary intracranial meningioma in the patients at and under 5 years old who received radiotherapy was faster than the development of such tumors in those who received radiotherapy at older ages. This period of time for development of the tumor was 16.1 [55, 64].

Many studies explaining the relationship between Hodgkin lymphoma and second primary tumors report that the risks of leukemia, breast cancer, thyroid cancer, sarcoma, and lung cancer are increased [65-74]. Regarding the central nervous system, spinal cord glioma after treatment for Hodgkin lymphoma has been reported in 3 cases in the literature [56, 59, 61]. Similarly, spinal meningioma was observed in one case [60]. The approximate period of occurrence of those cases after the treatment for Hodgkin lymphoma varied between 6 and 9 years after radiotherapy [60, 61]. The retrospective study performed by Behringer et al. with 5367 patients with Hodgkin lymphoma showed that

<table>
<thead>
<tr>
<th>Case</th>
<th>Tumor type</th>
<th>Age at radiation (year)</th>
<th>Radiation dose</th>
<th>Latency period (year)</th>
<th>Tumor location</th>
<th>Follow-up period</th>
<th>Author (year)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Glioblastoma multiforme</td>
<td>21</td>
<td>50</td>
<td>6</td>
<td>Cervico-thoracic</td>
<td>10 weeks&lt;sup&gt;a&lt;/sup&gt;</td>
<td>Clifton et al. 1980 [56]</td>
</tr>
<tr>
<td>2</td>
<td>Undifferentiated neurogenic tumor</td>
<td>3</td>
<td>NS</td>
<td>6</td>
<td>cerebral</td>
<td>NS</td>
<td>Schmitt et al. 1984 [57]</td>
</tr>
<tr>
<td>3</td>
<td>Gliosarcoma</td>
<td>6</td>
<td>NS</td>
<td>NS</td>
<td>cerebral</td>
<td>NS</td>
<td>Takaue et al. 1986 [58]</td>
</tr>
<tr>
<td>4</td>
<td>Astrocytoma grade II-III</td>
<td>19</td>
<td>40</td>
<td>7</td>
<td>Cervical</td>
<td>6 month</td>
<td>Bazen et al. 1990 [59]</td>
</tr>
<tr>
<td>5</td>
<td>Meningioma</td>
<td>15</td>
<td>140</td>
<td>27</td>
<td>Posterior fossa</td>
<td>3 year</td>
<td>Deutsch et al. 1999 [36]</td>
</tr>
<tr>
<td>6</td>
<td>Meningioma</td>
<td>26</td>
<td>NS</td>
<td>9</td>
<td>Cervical</td>
<td>NS</td>
<td>Martin et al. 2001 [60]</td>
</tr>
<tr>
<td>7</td>
<td>Anaplastic glioma</td>
<td>30</td>
<td>40</td>
<td>9</td>
<td>Cervico-thoracic</td>
<td>11 month&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Riffaud et al. 2006 [61]</td>
</tr>
<tr>
<td>8</td>
<td>High grade astrocytoma</td>
<td>23</td>
<td>180</td>
<td>3</td>
<td>Conus medullaris</td>
<td>Several weeks&lt;sup&gt;a&lt;/sup&gt;</td>
<td>Ng et al. 2007 [62]</td>
</tr>
<tr>
<td>9</td>
<td>Meningioma</td>
<td>4</td>
<td>25</td>
<td>6</td>
<td>Temporo-parietal</td>
<td>6 month</td>
<td>Present case</td>
</tr>
</tbody>
</table>

<sup>a</sup>exitus date, NS: not stated.

Table 1: CNS Tumors After Radiotherapy for Hodgkin Lymphoma in Literature
lung, colorectal and breast cancers were the most common after a 72-month follow-up. The risk of developing a secondary solid tumor was 2%. Of the patients who developed secondary tumors, 77.2% were treated with a combined treatment protocol [55].

Hodgkin’s lymphoma is common in underdeveloped areas and low socioeconomic status. The risk of Hodgkin lymphoma makes a peak in early childhood and the 3rd decade of life. Also risk increases with increased age [75]. Among tumors occurring during childhood, meningioma is very rare. Children accounted for only 1.9% of patients in the study of Cushing and Eisenhardt, which was performed with 313 patients with meningioma [76]. The frequency of meningioma in children was found to be approximately 2% in later studies [24]. These tumors account for 1-4.2% of all primary intracranial tumors during childhood [17-28].

Chordoid meningiomas, which are grade 2 according to the WHO classification, account for only

Table 2: Reported Cases of Intraparenchymal Meningioma in Child

<table>
<thead>
<tr>
<th>#</th>
<th>Age, Sex</th>
<th>Etiology</th>
<th>Location</th>
<th>Operation</th>
<th>Histology</th>
<th>Follow up</th>
<th>Author (year)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>5 mos, F</td>
<td>Primary</td>
<td>Frontal</td>
<td>Total</td>
<td>Fibroblastic</td>
<td>NS</td>
<td>Suematsu et al. 1974[28]</td>
</tr>
<tr>
<td>2</td>
<td>0-14 yrs, NS</td>
<td>Primary</td>
<td>NS</td>
<td>Craniotomy</td>
<td>Sarcomatous</td>
<td>NS</td>
<td>Sano et al. 1981 [79]</td>
</tr>
<tr>
<td>3</td>
<td>0-14 yrs, NS</td>
<td>Primary</td>
<td>NS</td>
<td>Craniotomy</td>
<td>Fibroblastic</td>
<td>NS</td>
<td>Sano et al. 1981 [79]</td>
</tr>
<tr>
<td>4</td>
<td>1 yrs, M</td>
<td>Primary</td>
<td>Parietal</td>
<td>Total</td>
<td>Fibroblastic</td>
<td>NS</td>
<td>Legius et al. 1985 [35]</td>
</tr>
<tr>
<td>5</td>
<td>12 yrs, M</td>
<td>Primary</td>
<td>Temporal</td>
<td>Partial</td>
<td>Transitional</td>
<td>3 yrs</td>
<td>Drake et al. 1986 [19]</td>
</tr>
<tr>
<td>6</td>
<td>7 yrs, M</td>
<td>Primary</td>
<td>Frontal</td>
<td>Total</td>
<td>Fibroblastic</td>
<td>NS</td>
<td>Schroeder et al. 1987 [80]</td>
</tr>
<tr>
<td>7</td>
<td>11 mos, M</td>
<td>Primary</td>
<td>Frontal</td>
<td>Total</td>
<td>Fibroblastic</td>
<td>5 yrs</td>
<td>Kimura et al. 1987 [81]</td>
</tr>
<tr>
<td>8</td>
<td>2 yrs, F</td>
<td>Primary</td>
<td>Frontal</td>
<td>Total</td>
<td>Psammo-matous</td>
<td>4 mos</td>
<td>Mamourian et al. 1991 [82]</td>
</tr>
<tr>
<td>9</td>
<td>6 yrs, F</td>
<td>Primary</td>
<td>Frontal</td>
<td>Total</td>
<td>Transitional</td>
<td>2 yrs</td>
<td>Matsumoto et al. 1992 [83]</td>
</tr>
<tr>
<td>10</td>
<td>12 yrs, F</td>
<td>Primary</td>
<td>Temporal</td>
<td>Total</td>
<td>Meningo-thelial</td>
<td>NS</td>
<td>Kaneko et al. 1993 [84]</td>
</tr>
<tr>
<td>11</td>
<td>2 yrs, M</td>
<td>Primary</td>
<td>Temporal</td>
<td>Craniotomy</td>
<td>Fibroblastic</td>
<td>NS</td>
<td>Miwa et al. 1996 [85]</td>
</tr>
<tr>
<td>12</td>
<td>1 yrs, F</td>
<td>Primary</td>
<td>Frontal</td>
<td>Total</td>
<td>Fibroblastic</td>
<td>2 yrs</td>
<td>Kohama et al. 1996 [34]</td>
</tr>
<tr>
<td>13</td>
<td>2 yrs, F</td>
<td>Primary</td>
<td>Brain stem</td>
<td>Partial</td>
<td>Clear cell</td>
<td>NS</td>
<td>Teo et al. 1998 [86]</td>
</tr>
<tr>
<td>14</td>
<td>14 yrs, M</td>
<td>Primary</td>
<td>Temporal</td>
<td>Total</td>
<td>Fibroblastic</td>
<td>3 yrs</td>
<td>Karadereler et al. 2004 [87]</td>
</tr>
<tr>
<td>15</td>
<td>16 yrs, M</td>
<td>Primary</td>
<td>Occipital</td>
<td>Total</td>
<td>Atypical</td>
<td>1 yrs 6mos</td>
<td>Zhang et al. 2007 [88]</td>
</tr>
<tr>
<td>16</td>
<td>10 yrs, M</td>
<td>Primary</td>
<td>Frontal</td>
<td>Total</td>
<td>Meningo-thelial</td>
<td>5 mos</td>
<td>Shimbo et al. 2011 [77]</td>
</tr>
<tr>
<td>17</td>
<td>10 yrs, M</td>
<td>Secondary</td>
<td>Temporoparietal</td>
<td>Total</td>
<td>Chordoid</td>
<td>6 mos</td>
<td>Present case</td>
</tr>
</tbody>
</table>

0.5-1% of all meningiomas [30]. As with all meningiomas, the chordoid form is common in adults and very rare in children [26, 64].

In a review of the literature describing meningiomas during childhood that were histopathologically examined, the frequency of grade 2 meningioma varied between 2% and 5%. In a series of 38 patients, grade 2 meningioma accounted for 21.9%, and only 4.9% (2 of 38 patients) had chordoid meningioma pathology [24]. During childhood, convexity and parasagittal meningiomas are the most common, followed by meningiomas at the skull base and the posterior fossa [24]. Intraparenchymal or subcortical meningiomas without dural connections are even rarer [29]. There are only 24 cases of intraparenchymal meningioma reported in the literature [77]. Of these, only one was a chordoid meningioma [78]. The cases of intraparenchymal meningiomas during childhood that have been published in the literature are presented in Table 2.

The term ‘chordoid meningeal tumor’ was used by Kepes et al. for the first time in 1988 [89]. Chordoid meningioma is a rare variant of meningioma and accounts for 0.5-2% of all meningiomas [30, 31, 90, 91]. Chordoid meningioma has a great risk of recurrence and aggressive growth [92-94] and is classified as grade 2 according to WHO [92]. To date, 53 cases have been published in the literature, and only 6 of these patients were children [26, 31, 95, 96]. Of these 53 tumors, 4 tumors were located in the infratentorial region and 11 in the supratentorial region. A large proportion of supratentorially located tumor are localized around the falx cerebri in the convexity [95]. Intraparenchymal chordoid meningioma has not been published in the literature.

In our case, the secondary meningioma was located in the parenchyma without dural connections. The presented case is the second case of intraparenchymal chordoid meningioma in the literature and is the only case in a child.

Table 3: Radiation Induced Meningioma with Short Latency Period

<table>
<thead>
<tr>
<th>Case</th>
<th>Age, Sex</th>
<th>Primary Malignancy</th>
<th>Cranial radiation TD/fr (cGy)</th>
<th>Latency Period (months)</th>
<th>Location</th>
<th>Histology</th>
<th>Follow up</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>4, F</td>
<td>Optic nerve glioma</td>
<td>65</td>
<td>48</td>
<td>Basifrontal</td>
<td>Anaplastic meningioma</td>
<td>1 year</td>
</tr>
<tr>
<td>2</td>
<td>4, M</td>
<td>Medullo-blastoma</td>
<td>46</td>
<td>60</td>
<td>Right frontal</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>3</td>
<td>2,1 M</td>
<td>Left pontine glioma</td>
<td>55</td>
<td>42</td>
<td>Left Temporal</td>
<td>Benign meningioma</td>
<td>NS</td>
</tr>
<tr>
<td>4</td>
<td>??</td>
<td>Recurrent Pituitary Adenoma</td>
<td>HD</td>
<td>12</td>
<td>Skull base</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>5</td>
<td>7, M</td>
<td>Cranio-pharyngioma</td>
<td>50</td>
<td>24</td>
<td>Right temporal</td>
<td>Atypical meningioma</td>
<td>2 years</td>
</tr>
<tr>
<td>6</td>
<td>2, F</td>
<td>Medullo-blastoma</td>
<td>48.25</td>
<td>60</td>
<td>Convexity meningioma</td>
<td>Atypical meningioma</td>
<td>NS</td>
</tr>
<tr>
<td>7</td>
<td>4.5, M</td>
<td>Medullo-blastoma</td>
<td>HD</td>
<td>26</td>
<td>Parasagittal</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>8</td>
<td>9, F</td>
<td>Pineo-blastoma</td>
<td>HD</td>
<td>42</td>
<td>Parasagittal</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>9</td>
<td>&lt;3, ?</td>
<td>Medullo-blastoma</td>
<td>53.2</td>
<td>35</td>
<td>NS</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>10</td>
<td>9, M</td>
<td>Cutaneos angioma</td>
<td>25</td>
<td>60</td>
<td>Parietooccipital</td>
<td>Meningothelial</td>
<td>NS</td>
</tr>
<tr>
<td>11</td>
<td>11, M</td>
<td>Medullo-blastoma</td>
<td>54</td>
<td>14</td>
<td>Multiple skull base</td>
<td>Atypical meningioma</td>
<td>NS</td>
</tr>
<tr>
<td>12</td>
<td>4, M</td>
<td>Hodgkin lymphoma</td>
<td>25</td>
<td>60</td>
<td>Temporoparietal</td>
<td>Chordoid</td>
<td>6 month</td>
</tr>
</tbody>
</table>

F: female, M: male, HD: high dose, NS: not stated.
Radiation is a clear cause of meningioma. Meningiomas are the most common radiation-associated tumors of the central nervous system. Particularly in children, radiation exposure in high doses increases the risk of radiation-associated tumor development by 10-fold [97, 98]. The cases of radiation-associated meningioma during childhood after a short latent period in the early stage in the literature are listed in Table 3.

Radiation-associated meningiomas typically occur in the early ages. These tumors may primarily arise from the areas exposed to radiation and may be multifocal. The latent period is shorter in children than in adults [97]. In a study performed by Primoz et al., 170000 patients were retrospectively evaluated, and of these, 1227 were at or under 16 years old. Only 5 patients were diagnosed with meningiomas that met the criteria of radiation-associated tumors [55]. The ages of those patients varied from 3 to 13, and the patients received radiotherapy for different reasons. The first meningioma developed 9.5 years after radiotherapy and the average time is 19.7 years [55]. The risk of developing secondary intracranial meningioma was investigated in the same study and was found to be 0.53% for the first 10 years, 1.2% for 20 years and 8.18% for 25 years after radiotherapy [55]. The children who did not receive radiotherapy did not develop meningioma. Multiple meningiomas were found in the 60% of the patients who were diagnosed with meningioma [107], and only 20% of them [108] exhibited atypical meningioma pathology [55]. In the same study, there were 126 cases of radiation-associated meningioma, and of these, 68% involved radiotherapy during childhood. The latent period was shorter in children, although there was no significant relationship between the age of radiotherapy and the latent period; the average latent period is 16.1 years [55].

In the case presented herein, the patient, who received radiotherapy after a diagnosis of Hodgkin lymphoma, developed meningioma in a very short period of time, in contrast to other cases reported in the literature. After a rare pathology, this tumor developed into a very rare histopathological type in an unusual location, thus representing a rare complication. The radiotherapy-associated intraparenchymal chordoid meningioma was totally excised. The patient was stable in terms of his reaction to combined radiotherapy and chemotherapy and to the status of his Hodgkin lymphoma, and his intracranial pathology was immunohistochemically examined. Immunohistochemical examinations are used primarily to define meningiomas. MIB-1 labeling, which is considered to be closely related to the histological grading of meningiomas, is a technique that can be used to visualize proliferative activity [109-111]. The MIB-1 index is a good tool to evaluate the risk of malignancy and recurrence [112-114]. There are studies in the literature showing that the MIB-1 index has higher values in atypical and malignant meningiomas than in typical meningiomas [113, 115, 118]. It is stated in the literature that a higher MIB-1 index is closely related with recurrence [113, 115, 117-119]. The MIB-1 index was 10 % in our case. This level indicates a high risk of recurrence, and therefore, this patient must be monitored closely.

The negative prognostic factors in meningioma are as follows: a) atypical or anaplastic histological stage, b) aggressive histological variant (clear cell, chordoid, papillary, rhabdoid), c) young patient (< 40 years), d) high MIB-1 index, e) affected anterior visual path, and f) genetic changes related to progression [95]. The MIB-1 index is approximately 3.8 in benign meningiomas, 7.2 in atypical meningiomas and 14.7 in anaplastic meningiomas [95]. In a study performed by Couce et al., after a long-term follow-up, one or more recurrences were observed in 39% of patients with meningiomas, with the recurrences occurring 5.6 years later on average [120]. All of the patients underwent subtotal resection except for one patient with recurrence. A total of 87.7% of relapsed patients had over 50% chordoid patterns [120].

Our patient is included in the high risk group for recurrence due to the histological stage, the chordoid tumor type, his age, the tumor’s high MIB-1 index. The total excision of intracranial lesion reduces the risk of recurrence.

**CONCLUSION**

Although Hodgkin lymphoma is a disease that can be treated with radiotherapy and/or chemotherapy in every stage, the therapy-associated secondary tumors that can occur in the late stage are the primary cause of mortality and morbidity for patients with Hodgkin lymphoma. Surgeons must remember that radiation-associated meningiomas may occur in the early stage of the treatment as well as in the late stage. Young patients with grade 2 chordoid meningiomas should be followed up in case of recurrence, and those with higher Ki-67 values are highly expected to relapse.
REFERENCES


Radiation-Associated Intraparenchymal Meningioma

Journal of Cancer Research Updates, 2014 Vol. 3, No. 1


Journal of Cancer Research Updates, 2014 Vol. 3, No. 1

http://dx.doi.org/10.1007/s11060-005-5532-y


Received on 03-08-2013 Accepted on 27-11-2013 Published on 13-02-2014

DOI: http://dx.doi.org/10.6000/1929-2279.2014.03.01.4

© 2014 Efendioglu et al.; Licensee Lifescience Global. This is an open access article licensed under the terms of the Creative Commons Attribution Non-Commercial License (http://creativecommons.org/licenses/by-nc/3.0/) which permits unrestricted, non-commercial use, distribution and reproduction in any medium, provided the work is properly cited.