Cancer/Radiothérapie 21 (2017) 261–266

Disponible en ligne sur
ScienceDirect
www.sciencedirect.com

Elsevier Masson France
EM|consulte
www.em-consulte.com

Original article

Primary spinal glioma in children: Results from a referral pediatric institution in Shanghai

Gliome de la moelle épinière chez l'enfant : résultats d'une série de 11 cas à Shanghai

J. Khalil[a], Z. Chuanying[b], Z. Qing[b], Y. Belkacémi[c], J. Mawei[b,*]

[a] *Radiation oncology, National Cancer Institute, Souissi, 10000 Rabat, Morocco*
[b] *Radiotherapy, Xhinhua General Hospital, 1665 Kongjiang Road, Yangpu, 10000 Shanghai, China*
[c] *Radiotherapy, hôpital Henri-Mondor, avenue du Maréchal-de-Lattre-de-Tassigny, 94010 Créteil cedex, France*

ARTICLE INFO

Article history:
Received 4 September 2016
Received in revised form 9 November 2016
Accepted 17 November 2016

Keywords:
Primary glioma
Spinal
Children

ABSTRACT

Purpose. – Primary spinal cord glioma is a rare entity especially in children; accounting for less than 10% of all central nervous system tumors. Low grade is the most reported subtype. Treatment modalities have largely evolved; large improvements have been made in the surgical field but also in both of radiotherapy and chemotherapy. Nevertheless, the optimal treatment is yet to be defined.
Material and methods. – A chart review of 11 pediatric patients with a diagnosis of low grade spinal cord glioma at Xhinhua hospital in Shanghai was conducted. A statistical package for Social Sciences Package (SPSS) was used for analysis. Means and standard deviations were calculated. The Kaplan-Meier method was used to analyze overall survival and progression-free survival.
Results. – The mean age was 6.7 years (range: 6 months–14.3 years). Revealing symptoms were variable and slowly progressive. The mean duration of symptoms prior to diagnosis was of 7 ± 3.2 months. Astrocytoma was the most commonly reported histological type (seven cases, 63.6%), ependymomas were reported in three cases (27.3%). Surgery was performed in all patients. Subtotal resection concerned the majority of patients (nine patients, 81.8%). Adjuvant radiotherapy was indicated in all cases. A total dose of 39.6 Gy was delivered to the whole group. Three patients received adjuvant chemotherapy, of whom two patients had grade III glioma and one patient had a tumor recurrence. Temozolomide-based regimen was the main protocol used for all our patients. The 3 years overall survival rate was 100%, whereas the progression free survival rate was 87.5%. One case relapsed during the next year following completion of treatment.
Conclusion. – Our preliminary results are consistent with that of other similar published reports, however longer follow up is needed. So are specific recommendations that are still lacking in this setting.

Mots clés :
Gliome
Moelle épinière
Enfant

RÉSUMÉ

Objectif de l'étude. – Le gliome de la moelle épinière est une entité rare, surtout chez les enfants, représentant moins de 10 % de toutes les tumeurs du système nerveux central. Le bas grade est le sous-type le plus fréquent. Les modalités de traitement ont largement évolué. De grandes améliorations ont été apportées dans le domaine chirurgical, mais aussi en radiothérapie et en chimiothérapie. Néanmoins, le traitement optimal doit encore être défini.
Matériel et méthodes. – Une revue des dossiers de 11 enfants atteints de gliome de moelle épinière de bas grade à l'hôpital Xhinhua de Shanghai a été mené. Le logiciel SPSS a été utilisé pour l'analyse. Les moyennes et les écarts types ont été calculés. La méthode de Kaplan-Meier a été utilisée pour analyser la survie globale et la survie sans progression.

* Corresponding author.
E-mail address: bensaidbadr8@gmail.com (J. Mawei).

http://dx.doi.org/10.1016/j.canrad.2016.11.005

Résultats. – L'âge moyen était de 6,7 ans (6 mois–14,3 ans). Les symptômes révélateurs étaient variables et lentement progressifs. La durée moyenne des symptômes avant le diagnostic était de 7 ± 3,2 mois. Les astrocytomes étaient le type histologique le plus fréquent (sept cas, 63,6 %), puis les épendymomes (trois cas, 27,3 %). La chirurgie a été réalisée chez tous les patients, subtotale chez la majorité (neuf patients, 81,8 %). Une radiothérapie adjuvante a été indiquée dans tous les cas. Une dose totale de 39,6 Gy a été délivrée. Trois patients ont reçu une chimiothérapie adjuvante, dont deux étaient atteints d'un gliome de grade III et un d'une récidive tumorale. Le témozolomide a été utilisé pour tous les patients. La probabilité de survie globale de 3 ans était de 100 %, alors que celle de survie sans progression était de 87,5 %. Un gliome a rechuté l'année après l'achèvement du traitement.
Conclusion. – Nos résultats préliminaires concordent avec ceux d'autres séries publiées, mais un suivi plus long est nécessaire. Il en va de même des recommandations, qui font encore défaut dans ce contexte.

1. Introduction

Primary spinal cord glioma is a rare entity accounting for less than 10% of all central nervous system tumors [1–3]. Of this, low grade is the most reported subtype and concerns 30 to 50% of the cases [4]. Revealing symptoms are mostly progressive sensorimotor and autonomic deficits occurring secondary to the involvement of multiple tracts within the spinal cord. These past few years, treatment modalities have largely evolved; in fact advances in imaging techniques have led to an improvement in surgical treatment allowing more complete resections [5–7]. These advances concerned not only the surgical field but also radiotherapy; the role of postoperative radiotherapy using the newest techniques has largely matured. Nevertheless, given the small-published series, neither technical aspects of radiotherapy nor treatments' guidelines are clearly established [8–14]. Prognostic factors impacting treatments' outcomes are also divergent in the available data of the literature [15,16]. As our institution is the referral center for pediatric tumors in Shanghai, we reviewed the data of pediatric patients diagnosed with primary spinal cord gliomas and treated in Xinhua hospital. Accordingly, we will report our experience in the treatment of this rare entity and provide the literature with one more series analyzing overall survival, progression-free survivals, and prognostic factors of this patient population, in an attempt to draw some conclusions regarding the best treatment approaches for this patient population.

2. Patients and methods

Our study is a retrospective one that concerned pediatric patients diagnosed with spinal cord low-grade glioma. Charts were obtained from the medical records department at the Xhinhua hospital in Shanghai, China from 2009 to 2013. No patient was excluded from the series. The following data were obtained from the patients' chart: Age, sex, date of birth, signs and symptoms, length of symptoms before diagnosis, diagnostic imaging tool. The location, histology of the tumor, histological subtype, grade and the quality of surgical resection (complete or incomplete) were recorded. Radiation and chemotherapy protocols, follow-up neuroimaging, metastatic work-up, treatment related toxicities and treatment management for relapses were also reviewed.

3. Statistical analysis

A Statistical Package for Social Sciences package (SPSS) version 18 was used for analysis. Means and standard deviations were calculated. The Kaplan-Meier method was used to analyze overall and progression-free survival.

Table 1
Spinal cord glioma in paediatric patients, study on an 11 patient cohort in Shanghai: Clinicopathologic criteria.
Gliome de la moelle épinière chez l'enfant, étude d'une cohorte de 11 patients à Shanghaï : critères clinicopathologiques de la cohorte.

Criteria	*n* (%)
Age	Mean age: 6.7 years (range: 6 months–14.3 years)
Sex	
Boy	9 (81.8%)
Girl	2 (11.8%)
Focal neurology symptoms	
paraparesis	4 (44%)
monoparesis	2 (18.2%)
sphincter problems	3 (27.3%)
sensory problems	1 (9%)
Imaging modalities	
MRI	11 (100%)
CT	0
Physical exam	
Tendon reflex changes	3 (33%)
Motor deficits	2 (18.2%)
Neurologic status (McCormick scale)	
Grades I and II	8 (72.7%)
Grades III and V	3 (27.3%)
Tumor location	
cervical spine	4 (36.3%)
Thoracic spine	5 (45.4%)
Lumbar spine	4 (36.3%)
Number of segments	
< 5	3 (27.3%)
> 5	8 (72.7%)

4. Results

Between January 2009 and December 2013, 11 children were diagnosed with primary spinal glioma at Xhinhua hospital and were accordingly included in the study.

4.1. Clinicopathological criteria

The mean age for the group of patients was 6.7 years (range: 6 months–14.3 years), and the median age was 5.7 years. The male/female ratio was 4.5. Clinicopathological criteria of the studied cohort are summarized in Table 1.

Revealing symptoms were variable and nonspecific. Young patients ($n = 3$, 27.3%) presented with nonspecific crying episodes followed by motor weakness and torticollis. Older patients ($n = 8$, 72.6%) presented with back pain, progressive kyphoscoliosis, and motor dysfunction. Focal neurology signs were reported in six cases, it consisted on paraparesis (four patients), monoparesis (two patients), sphincter problems (three patients), and sensory problems (one patient). A palpable mass was the presenting symptom