

# Gamma knife surgery-induced ependymoma after the treatment of meningioma – a case report

## *Wyściółczak wywołany leczeniem oponiaka za pomocą noża gamma – opis przypadku*

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Neurologia i Neurochirurgia Polska 2012; 46, 3: 294-296

DOI: 10.5114/ninp.2012.29138

### Abstract

Gamma knife surgery is widely used for a number of neurological disorders. However, little is known about its long-term complications such as carcinogenic risks.

Here, we present a case of a radiosurgery-induced ependymoma by gamma knife surgery for the treatment of a spinal meningioma in a 7-year-old patient. In light of reviewing the previous reports, we advocate high caution in making young patients receive this treatment.

**Key words:** gamma knife, radiation-induced tumor, ependymoma.

### Introduction

Nowadays, gamma knife surgery plays an important role in the therapeutic strategy for various kinds of central nervous system disorders such as vascular malformations and tumours. In contrast to conventional radiotherapy, gamma knife surgery has the advantage of concentrating a high dose of radiation to an imaging-defined volume while the surrounding tissues only re-

### Streszczenie

Leczenie za pomocą noża gamma jest stosowane w wielu schorzeniach układu nerwowego. Niewiele jednak wiadomo o odległych powikłaniach tej terapii, np. o ryzyku wywołania wtórnych nowotworów.

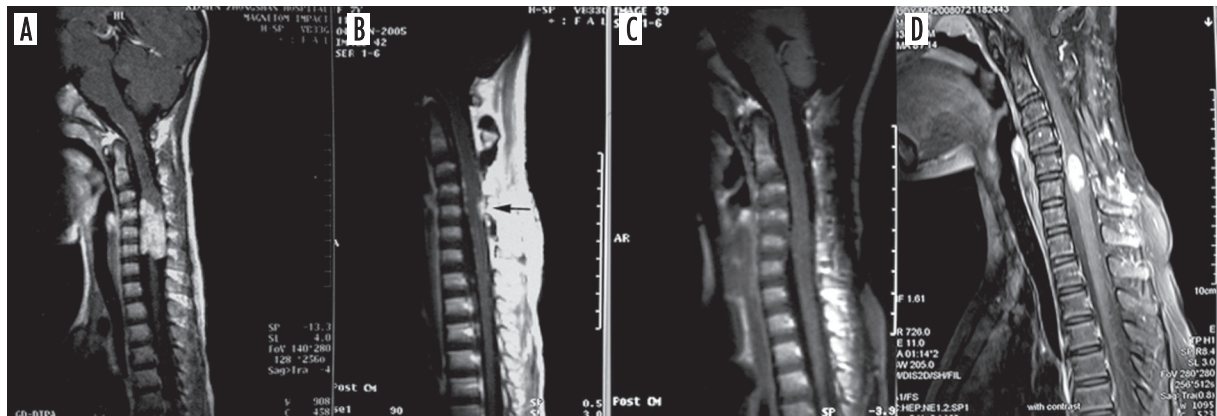
W pracy przedstawiono przypadek 7-letniej dziewczynki, u której leczenie oponiaka kanału kręgowego za pomocą noża gamma wywołało wyściółczaka wtórnego do leczenia radiochirurgicznego. W świetle wcześniejszych doniesień na ten temat autorzy uważają, że należy zachować dużą ostrożność w podejmowaniu tego rodzaju leczenia u pacjentów w młodym wieku.

**Słowa kluczowe:** nóż gamma, guz wywołany napromienianiem, wyściółczak.

ceive a very low dose of radiation. Since its introduction to clinical use, serious complications of gamma knife surgery are rarely observed, which may be due to a very low incidence rate and a relatively long latency period after radiosurgery. Carcinogenic risk of radiation therapy is a major concern for patients who decide whether to receive this treatment. Although this risk has been well documented for conventional radiotherapy, there is very limited knowledge about the risk of gamma knife surgery.

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Received: 10.03.2011; accepted: 5.10.2011

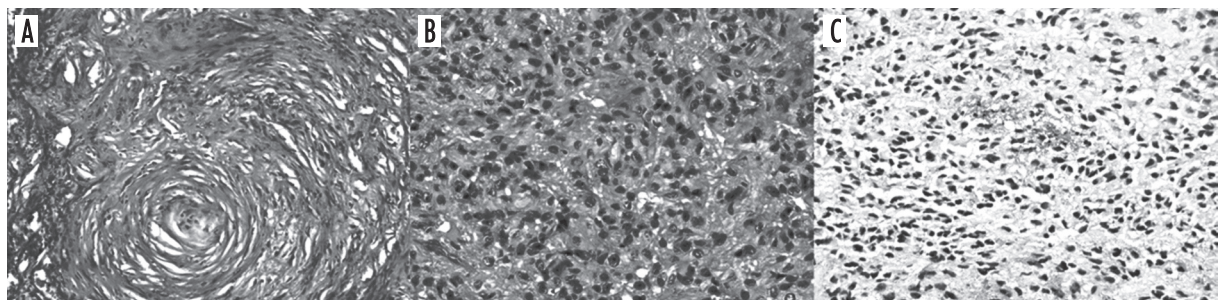


**Fig. 1.** Sagittal gadolinium-enhanced T1-weighted MR images: A) delineating the C4-C6 spinal tumour before the first operation; B) showing the spinal tumour residue before gamma knife surgery, indicated by the black arrow; C) no evidence of secondary tumour induction 7 months after gamma knife surgery; D) showing intramedullary lesion 3 years after gamma knife surgery

## Case report

In May 2005, this 7-year-old girl came to the local hospital because she suffered from weakness of her right arm. In the neurological examination, the muscle strength of her right upper limb decreased to grade IV. Magnetic resonance imaging (MRI) revealed a mass located at the cervical vertebral levels from C4 to C6 (Fig. 1A). She received the first operation at the local hospital and a subtotal removal of the lesion was obtained. After the operation, the pathology turned out to be a meningioma (WHO grade I) (Fig. 2A). The girl recovered well and the muscle strength of her right arm gradually improved to grade V. The MRI scan performed 9 days after the operation showed that a small part of the tumor remained (Fig. 1B). One month later, in July 2005, the neurosurgeons at the local hospital arranged for her to undergo gamma knife surgery for the residual tumor with a marginal dose of 20 Gy at the 50% isodose line. She went through the radiosurgery well and follow-up by serial

MRIs was suggested. The MRI scan taken 7 months after radiosurgery showed that the meningioma residue had diminished and there was no sign of a secondary lesion (Fig. 1C). After that, the patient experienced a symptom-free period until July 2008 when, at the age of 10, she presented with right arm weakness again. She went to the local hospital and received another MRI, which showed an intramedullary lesion located at the C5/C6 vertebral level (Fig. 1D). This time the patient was referred to our department for the second operation. During the surgery, we confirmed that the mass was located inside the spine tissue, and we found there was no extramedullary mass, which was indicative of no recurrence of the primary meningioma. Then we succeeded in performing a gross total resection of the intramedullary mass. The pathology of the intramedullary mass was diagnosed to be an ependymoma (WHO grade II) (Figs. 2B, 2C). Postoperatively, her right arm regained its strength little by little, and we ordered serial follow-up MRIs for the patient.



**Fig. 2.** Photomicrographs of surgically resected tumour tissue sections by histopathological and immunohistochemical staining. The tumour tissue from the first operation stained with haematoxylin-eosin (H&E) showed the typical structure of whorls which consisted of elongated spindle cells ( $\times 100$ ) (A); the tumour tissue from the second operation showed the aggregates of ependymocytes stained with H&E ( $\times 200$ ) (B) and a portion of GFAP-positive tumor cells ( $\times 200$ ) (C)

## Discussion

Due to its noninvasiveness and low morbidity rate, stereotactic radiosurgery such as gamma knife surgery has become a very useful therapeutic tool to manage a myriad of disorders in the central nervous system. In the management of central nervous system (CNS) tumours, gamma knife surgery can serve as an adjuvant therapeutic strategy after subtotal removal or partial removal of a tumour, or it can even be the treatment of choice on some occasions when surgical intervention becomes too risky for patients. With the widespread use of gamma knife surgery, both therapists and patients are becoming concerned about its long-term complications, such as the possibility of tumour induction. Because of the technique's superiority, normal tissues receive a lower dose of radiation in patients treated by gamma knife surgery than in those treated by conventional radiotherapy. Therefore, patients undergoing gamma knife surgery are expected to face a lower incidence rate of tumor induction than those receiving conventional radiotherapy. The estimated risk of radiation-induced tumor after conventional radiotherapy to the CNS is approximately 1-3% [1]. So far, only six cases of brain tumours induced by gamma knife surgery have been reported, including four cases of glioblastoma multiforme, one case of meningioma, and one case of anaplastic astrocytoma [1-6]. All these tumours occurred inside the cranium, and no spinal tumour has been reported to be induced by gamma knife surgery. We have reported the first case in which a spinal ependymoma was induced by gamma knife surgery for the treatment of the residue of a spinal meningioma.

According to the definition made by Cahan *et al.* [7], a radiation-induced tumour should meet the following criteria: 1) the tumour occurs within the field of irradiation used to treat the primary disease; 2) the tumour is not present prior to irradiation, and there must be a latency interval between radiotherapy and the detection of the second tumour; 3) the secondary tumour must be histologically distinct from the primary disease; 4) the patient must not have genetic predisposing conditions for cancer development. Apparently, our case fulfils these criteria. We do not believe this case was a change in tumour type because the primary tumour and secondary tumour originated from different areas: extramedullary versus intramedullary. Nor do we think this case occurred as an incidental combination of meningioma and ependymoma because the secondary neoplasm showed up 3 years after radiosurgery. The follow-up MRI scan performed

7 months after gamma knife surgery showed no sign of the occurrence of a secondary neoplasm.

Notably, the latency interval in this case is about 3 years, which is much shorter than the latency intervals reported in the previous 6 cases. One reason may be the younger age of the patient in this case. A study was conducted to estimate the carcinogenic risk of conventional radiotherapy in children and adolescents, which concluded that an increased incidence of radiation-induced brain tumor could be expected in childhood and the latency interval varied from months to years [8]. Our case supports that this rule may also apply for children after stereotactic radiosurgery. Therefore, one should be very cautious about making young patients receive stereotactic radiosurgery, especially when this strategy is designed for the treatment of benign CNS lesions. Both neurosurgeons and patients need to be aware of the possibility of developing a radiosurgery-induced tumour. Considering her sensitivity to radiation and the gross total removal of the gamma knife surgery-induced ependymoma, we ordered close follow-up MRIs for the patient in this case.

## Disclosure

Authors report no conflict of interest.

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