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Clinical case

Long-term stabilization by radiosurgery of a secondary focal anaplastic transformation in a surgically treated WHO grade II oligodendroglioma. A case report

Stabilisation à long terme par la radiochirurgie d'un foyer de transformation anaplasique d'un oligodendrogliome de bas grade préalablement opéré

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ABSTRACT

We report on a young woman with a left temporal diffuse low-grade glioma treated initially by a subtotal resection. A focal anaplastic area appeared 5 years later and was treated by radiosurgery. A long-time stabilization was therefore obtained and lasted even after pregnancy, which is a known factor of faster tumour progression. This report shows that radiosurgery could be an option in the multimodal treatment of a selected group of patients with focal malignant transformation of diffuse low-grade glioma. It could permit long-term stabilization of the tumour without any other adjuvant treatment and without compromising the quality of life.

Mots clés :
Radiochirurgie
Survie à long terme
Oligodendrogliome anaplasique
Grossesse

RÉSUMÉ

Dans cet article, nous rapportons le cas d'une jeune patiente atteinte d'un gliome diffus de bas grade localisé dans le lobe temporal gauche, initialement traité par résection chirurgicale subtotale. Cinq ans après cette intervention, un foyer de transformation anaplasique apparut et fut traité par radiochirurgie. Ce traitement a permis au dernier suivi, sept ans après la radiochirurgie, une stabilisation tumorale malgré une maternité, facteur connu pour accélérer la vitesse de croissance de ces tumeurs. Cette observation montre que la radiochirurgie pourrait être une option thérapeutique valide en cas de transformation maligne focale d'un gliome diffus de bas grade. Cela pourrait permettre une stabilisation durable de la tumeur sans recours à un autre traitement adjuvant, tout en maintenant la qualité de vie du patient.

1. Introduction

Diffuse supratentorial hemispheric low-grade gliomas (LGGs), i.e. World Health Organization (WHO) grade II gliomas in adults, are infiltrative tumours that invariably grow and progress to a higher grade, leading to neurological deterioration and death. Maximal surgical resection while preserving functional brain areas is the first treatment option. In the absence of unfavourable prognostic factors (older age, incomplete resection, neurological symptoms), postoperative clinical and MR-imaging follow-up without any adjuvant treatment is recommended [1]. After recurrence and anaplastic transformation, the treatment is less defined and varies between

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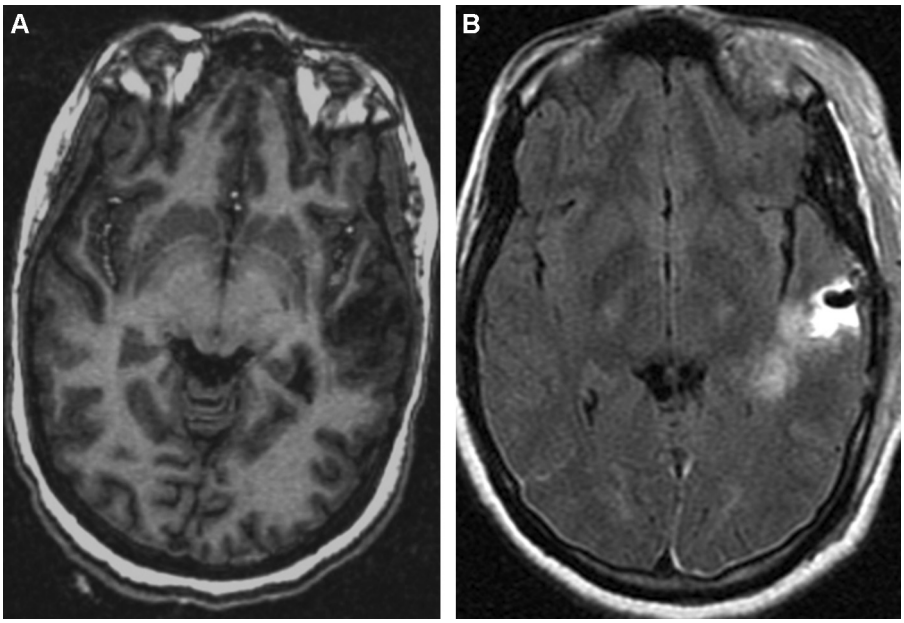

Fig. 1. Magnetic resonance imaging. A. Preoperative axial T1-weighted gadolinium-enhanced image showing a hypointense, non-enhanced left temporal lesion. B. Postoperative axial fluid-attenuated inversion-recovery-weighted image showing a small residual tumour in contact with the left ventricle.
Imagerie par résonance magnétique nucléaire. A. En préopératoire : coupe axiale en pondération T1 avec injection de gadolinium, montrant une lésion temporale gauche, hypointense et non rehaussée par le produit de contraste. B. En postopératoire : coupe axiale en pondération FLAIR, montrant un minime résidu tumoral au contacte du ventricule gauche.

new surgery, chemotherapy and/or radiotherapy. The role of radiosurgery is not well established but we hypothesize that it could permit stabilization of the tumour in selected groups of patients with focal relapse.

We present the first report of long-term stabilization by radiosurgery of a secondary focal anaplastic area in a supratentorial LGG initially treated by a subtotal resection. This stabilization lasted several years, even after pregnancy, which is a known risk factor of faster tumour progression [2].

2. Case report

A 17-year-old right-handed female without any previous medical history was admitted to the hospital after an inaugural generalized seizure. The neurological examination was normal except for some anomias (70/80 on the DO80 test). The brain MR-imaging showed a left temporal cortico-subcortical non-enhancing lesion evoking a diffuse LGG (Fig. 1A). This diagnosis was confirmed by biopsy, which also allowed us to exclude a dysembryoplastic neuroepithelial tumour, another possible diagnosis given the young age of the patient, the clinical presentation and anatomical location in the temporal lobe. A subtotal resection was then done in awake conditions according to functional boundaries. A small residue was voluntarily left in contact with the left sagittal stratum (Fig. 1B). As expected preoperatively, a right superior quadranopsia appeared. No other seizures occurred under antiepileptic drug therapy and the patient returned to a normal life. The examination of the entire tumour confirmed a WHO grade II oligodendroglioma and the immunohistochemical assessment found a Ki67 labelling index of 3%, the lack of expression of IDH1 and Internexine Alpha and a diffuse expression of Olig2. Only 1% of tumour cells expressed p53. MRI-based follow-up was assured without any adjuvant treatment. During the following years, a gradual increase of residual fluid-attenuated inversion-recovery (FLAIR) abnormalities occurred and a small (< 1 cm) nodular enhancing area appeared 5 years later (Fig. 2A). The progression of this area without increase of FLAIR abnormalities was confirmed by further MR-imaging done 6 weeks later. Due to the small volume of the enhanced part, it was not possible to perform good quality multimodal imaging. Focal anaplastic transformation was admitted without a new histological diagnosis. The decision was based on a previous study demonstrating that in the absence of any chemotherapy and/or radiotherapy the appearance of nodular enhancement in diffuse LGGs could be considered as focus of malignization [3]. Second surgery was rejected because of the location of the enhancing part, which was too close to the sagittal stratum and a radiosurgical treatment was then decided. Dynamic single-fractionated LINAC-based radiosurgery was performed using BRAINLAB stereotactic headframe. A non contrast-enhanced stereotactic CT-scan was done and a fusion with thin-slice gadolinium-enhanced T1-weighted MR-imaging obtained the day before was performed. The most aggressive, enhancing part of the lesion (diameter < 1 cm, volume 0.40 cm^3) was selected as a target volume. The maximal dose done on the center of the lesion was 23 Gy and a median marginal dose of 18.4 Gy was delivered to the 80% isodose line (Fig. 2B). The patient did not receive any corticoids and was discharged within 48 hours.

The clinical evolution was uneventful and after an initial increase, the enhancing tumour part completely disappeared about 2 years later. The FLAIR abnormalities also clearly shrank over time (Fig. 2C and D). The antiepileptic drug was stopped 1 year after radiosurgery and no new seizure occurred. The patient enjoyed a normal life and became pregnant 6 years later. Until the most recent MR-imaging performed more than 12 years after the diagnosis and 1 year after childbirth, the tumour remained completely stable.

3. Discussion

The management of diffuse supratentorial hemispheric LGGs represents one of the most challenging tasks for surgical and medical neuro-oncologists because of their frequent locations close to or within eloquent brain areas, the patients' young age and the usual lack of focal neurological deficit. Maximal safe