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Case report

SMART (stroke-like migraine attacks after radiation therapy) syndrome responded to steroid pulse therapy: Report of a case and review of the literature



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ARTICLE INFO	A B S T R A C T
Keywords: Glioma Radiation SMART syndrome Steroid pulse therapy	This report presents a case of stroke-like migraine attacks after radiation therapy (SMART) syndrome in a 31- year-old man in whom symptoms and radiological findings resolved with steroid pulsed therapy and reviews the literatures with special emphasis on the use of steroids against SMART syndrome. The patient had a past history of left temporal anaplastic astrocytoma and was treated with surgery followed by local 72 Gy radiation therapy and chemotherapy using Nimustine Hydrochloride. Four years after the surgery, he was suffering from subacute progressing symptoms of headache, right hemianopia, right hemiparesis and aphasia from 2 to 4 days before admission to our hospital. At first he was diagnosed as symptomatic epilepsy but after extensive examination, the final diagnosis was SMART syndrome. His symptoms soon improved with steroid pulse therapy. In the literature, steroid pulse therapy is not necessarily a standard of care for SMART syndrome, but it seemed to decrease the need of biopsy. As the lesions of SMART syndrome require differential diagnosis from recurrences, biopsy was performed in some cases. However, lack of benefit and possible detriment is reported with biopsy of SMART lesions. Through this experience we suggest that steroid pulse therapy may provide speedy recovery from symptoms, and it should be considered before other invasive investigations or treatments.

1. Introduction

SMART (stroke-like migraine attacks after radiation therapy) is a syndrome presumed to be a delayed complication of brain irradiation wherein patients have recurrent attacks of complex neurological symptoms, often including headache and seizures that begin many years after radiation therapy, and demonstrates characteristic imaging findings [1]. Since it was first described in 1995, about 40 cases of SMART have been reported in the literature [2]. Given the small number of cases reported to date, there is no clear consensus regarding effective treatment approaches to SMART.

2. Case presentation

A 31-year-old man, who had a past history of left temporal anaplastic astrocytoma, was then followed consistently with MRI at our outpatient department. He had initially been treated with surgery (Fig. 1, A, B) followed by local 72 Gy radiation therapy and chemotherapy using Nimustine Hydrochloride. After the treatment he had a right upper quadrantanopia but discharged without any other neurological deficit. He had received 12 cycles of maintenance ACNU therapy given once in 2 months and was then followed consistently. During this period, his symptomatic epilepsy was well controlled with levetiracetam and carbamazepine. Four years later, however, he suffered from subacute progressing symptoms of headache, right hemianopia, right hemiparesis and aphasia. MRI scan obtained on admission revealed abnormal findings, including new enhancement at left parietal lobe, and mild swelling of left frontal, parietal, and temporal gyri (Fig. 1, C, E). At first, he was treated with additional antiepileptic medication, because the electroencephalogram revealed focal slowing over the left frontal to temporal region. Nevertheless, his symptoms persisted for several days, especially his worsened visual field defect persisted for over 2 weeks. With the possible diagnosis of SMART, steroid pulse therapy (1000 mg/day, 5 days) was given. Subsequently, his visual field recovered to almost the similar level as before within only 2 weeks. Additionally, the abnormal imaging findings including enhancement and swelling had also disappeared on a subsequent MRI (Fig. 1, D, F). Clinical course of the present case is summarized in Fig. 1, G.

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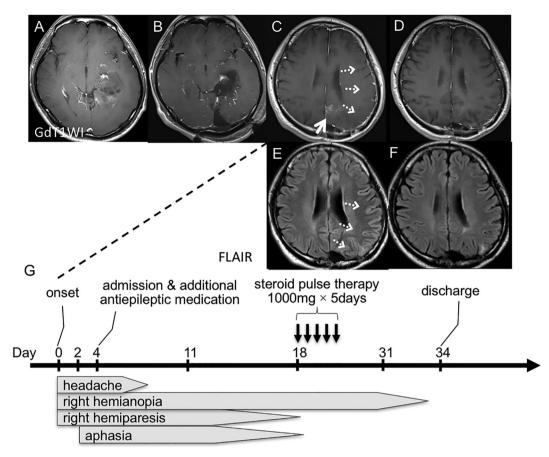


Fig. 1. Imaging findings with the clinical course.

Patient's preoperative T1-weighted image (T1WI) with contrast enhancement shows a left temporal anaplastic astrocytoma (A). Postoperative examination demonstrates removal of the lesion (B). T1WI with contrast enhancement and FLAIR (fluid attenuated inversion recovery) signal on admission revealed new enhancement at left parietal lobe (C, white arrow), and mild swelling of left frontal and temporal gyri (C, E, white dashed arrows). Three days after steroid pulse therapy, the enhancement and swelling had disappeared (D, F). Clinical course (G).

Table 1

Cases of SMART treated with steroids.

Author, year [ref]	Treatment	Recovery period after treatment	Outcome	Biopsy
Black, 2013 [4]	2 cases: corticosteroids	Undescribed	Partially recovered	1/2 biopsy
	1 case: antiepileptic drugs + corticosteroids	2.5 months	Completely recovered	-
	2 cases: antiepileptic drugs + varapamil + corticosteroids	2–2.5 months	Completely recovered	-
Di Stefano, 2013 [5]	1 case: antiepileptic drugs + methylprednisolone (1000 mg/day, 5 days)	4 days	Partially recovered	-
	3 cases: antiepileptic drugs + methylprednisolone (1000 mg/day, 5 days)	2–5 days	Completely recovered	-
	1 case: antiepileptic drugs + dexamethasone 20 mg daily	6 days	Partially recovered	-
Maloney, 2014 [6]	1 case: antiepileptic drugs + temporal lobectomy + dexamethasone (20 mg/day)	4 days	Completely recovered	-
Hametner,2015 [7]	1 case: antiepileptic drugs + high dose cortisone therapy + sedation	2 months	Completely recovered	-
Gómez-Cibeira, 2015 [1]	1 case: antiepileptic drugs + methylprednisolone (1000 mg/day, 5 days)	Several weeks	Completely recovered	-
Jaraba, 2015 [8]	1case: antiepileptic drugs + high dose corticosteroid therapy (1 mg/kg/ day) + sedation	Few days	Partially recovered but died of pneumonia	-
	1case: antiepileptic drugs + high dose corticosteroid therapy (1 mg/kg/ day)	1 week	Completely recovered	-
	1case: antiepileptic drugs + high dose corticosteroid therapy (1 mg/kg/ day)	11 days	Completely recovered	-
Nar Senol, 2015 [9]	1 case: antiepileptic drugs + dexamethasone	2 weeks	Completely recovered	-
Singh, 2016 [10]	1 case: antiepileptic drugs + corticosteroids	-	Permanent disability	-
Present case	1 case: antiepileptic drugs + steroid pulse therapy (1000 mg/day, 5 days)	2 weeks	Completely recovered	-

Ref; reference.

Steroid therapies and outcomes in reported SMART cases. In 5 cases who received steroid pulse therapy (1000 mg/day, 5 days), complete recovery was observed in 4 cases (80%) whereas only1 case (20%) just had partial recovery.

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