

Transarterial embolisation for refractory bilateral chronic subdural hematomas in a case with dentatorubral-pallidoluysian atrophy

Yoshihiro Tsukamoto · Makoto Oishi ·
Junnsuke Shinbo · Yukihiko Fujii

Received: 14 July 2010 / Accepted: 19 November 2010 / Published online: 2 December 2010
© Springer-Verlag 2010

Dear Editor,

We herein report a novel experience with refractory bilateral chronic subdural hematoma (CSDH), showing rapid progression despite three drainage surgeries, in which control was finally achieved by transarterial embolisation (TAE) of the middle meningeal artery (MMA), in a case with dentatorubral-pallidoluysian atrophy (DRPLA).

Case report

A 64-year-old man, receiving treatment for DRPLA, complained of progressive aphasia and right hemiparesis for 1 week, and finally, developed consciousness disturbance. The history of head trauma was unclear. Computed tomography (CT) showed bilateral CSDH (Fig. 1a). After bilateral continuous drainages for 1 day, clinical symptoms resolved completely, and CT showed definite decreases in the hematoma volume, but not complete closure of the subdural space. Three weeks later, we operated for

recurrence of CSDH on the left side with aphasia and right hemiparesis. Another 2 weeks later, we performed a third drainage surgery for bilateral recurrences of CSDH with thorough irrigation, with normal saline and steroid administration.

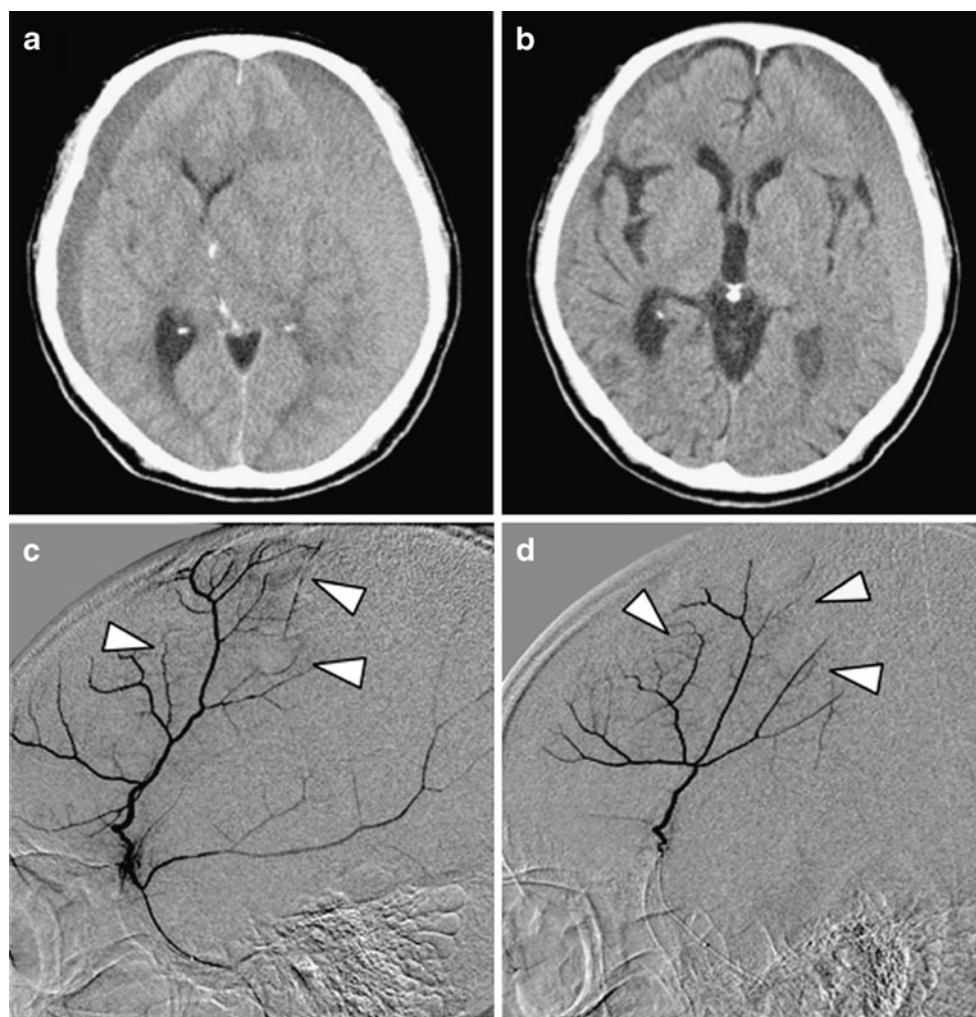
A further 2 weeks later, CT showed bilateral CSDH growth persistently (Fig. 1a), and then, we performed angiography to evaluate the underlying specific vascular diseases. Bilateral superselective angiography of the MMA showed diffuse abnormal vascular networks indicating the presence of macrocapillaries in the outer CSDH membrane (Fig. 1c, d). We performed TAE of the bilateral MMA with polyvinyl alcohol particles, and confirmed that the abnormal vessels were not visible. After 18 months since TAE of the MMA, the CT image still shows bilateral CSDH without enlargement, but the patient has no symptoms.

Discussion

Neurosurgeons sometimes encounter CSDH recurrence after drainage surgery, but the severe intractability of CSDH in the present patient is rarely reported [4, 6]. Burr hole drainage surgery is still the treatment of first choice for CSDH due to its effectiveness. However, when the CSDH recurs repeatedly, we select TAE of the MMA [3, 9, 10] as the second option prior to other therapeutic

Y. Tsukamoto (✉) · M. Oishi · J. Shinbo · Y. Fujii
Department of Neurosurgery, Brain Research Institute,
University of Niigata,
1-757 Asahimachi-dori, Chuo-ku,
Niigata 951-8585, Japan
e-mail: y-tsukamoto@bri.niigata-u.ac.jp

Fig. 1 CT images showing chronic subdural hematomas before the initial treatment (**a**) and at the third recurrence (**b**). Superselective angiographic images from frontal branches of the middle meningeal arteries via a microcatheter show stains (*arrowheads*) indicating abnormal vascular networks (**c** left side, **d** right side)



options, including Ommaya reservoir placement [7], subdural peritoneal shunting, and membranectomy via a large craniotomy. We are convinced of the high efficacy of this method based on the pathogenesis of CSDH. From the pathologic viewpoint, the outer CSDH membrane has macrocapillaries and shows invasion of macrophages and breakthrough bleeding of these macrocapillaries into the space at the dural border cell region. These changes are regarded as the fundamental cause of CSDH [5, 8]. The MMA is believed to affect CSDH growth by providing feeding vessels to the abnormal membrane surrounding the hematoma, which has a vascular connection with the dura matter. The angiographic findings of CSDH, presenting with diffuse MMA dilation and the abnormal vascular networks on its outer membrane, have been reported [9, 10]. The superselective MMA angiography in the present patient was consistent with these vascular findings.

We speculate that the intractability of CSDH in the patient may have been related to the brain condition of

DRPLA, a degenerative disease which causes not only the cerebellum and the brain stem but also the cerebrum to progressive atrophy. Previous authors indicated that reduced elasticity of the brain, rather than the brain atrophy itself, was possibly related to poor reexpansion of the brain, which can result in persistence of the postoperative subdural cavity as a risk factor for hematoma reaccumulation [2, 4, 7]. In that sense, DRPLA may be related to the reduced brain elasticity due to a decreasing cerebrovascular volume [1]. We believe that TAE of the MMA can be the best treatment option for refractory CSDH even with such underlying pathologies.

Acknowledgments Authors appreciate Drs. Masafumi Fukuda and Takatoshi Sorimachi for their clinical and technical assistances.

Conflicts of interest None.

References

1. Arai T, Mizukami K, Matsuzaka H, Iwakuma A, Shiraishi H, Koizumi J (1993) CNS changes in DRPLA with dementia and personality changes: CT, MR and SPECT findings. *Psychiatr Clin Neurosci* 47:105–110
2. Fukuhara T, Gotoh M, Asari A, Ohmoto T, Akioka T (1996) The relationship between brain surface elastance and brain reexpansion after evacuation of chronic subdural hematoma. *Surg Neurol* 45:570–574
3. Mandai S, Sakurai M, Matsumoto Y (2000) Middle meningeal artery embolization for refractory chronic subdural hematoma. Case report. *J Neurosurg* 93:686–688
4. Mori K, Maeda M (2001) Surgical treatment of chronic subdural hematoma in 500 consecutive cases: clinical characteristics, surgical outcome, complications, and recurrence rate. *Neurol Med Chir* 41:371–381
5. Murakami H, Hirose Y, Sagoh M, Shimizu K, Kojim M, Gotoh K, Mine Y, Hayashi T, Kawase T (2002) Why do chronic subdural hematomas continue to grow slowly and not coagulate? Role of thrombomodulin in the mechanism. *J Neurosurg* 96:877–884
6. Oishi M, Toyama M, Tamatani S, Kitazawa T, Saito M (2001) Clinical factors of recurrent chronic subdural hematoma. *Neurol Med Chir* 41:382–386
7. Sato M, Iwatsuki K, Akiyama C, Kumura E, Yoshimine T (2001) Implantation of a reservoir for refractory chronic subdural hematoma. *Neurosurgery* 48:1297–1301
8. Takahashi Y, Ohkura A, Yoshimura F, Ochiai S, Hirohata M, Shigemori M (1996) Ultrastructure of collagen fibers in the outer membrane of recurrent chronic subdural hematoma. *Neurol Med Chir* 36:627–630
9. Takahashi K, Muraoka K, Sugiura T, Maeda Y, Mandai S, Gohda Y, Kawachi M, Matsumoto Y (2002) Middle meningeal artery embolization for refractory chronic subdural hematoma: 3 case reports. *Noshinkeigeka* 30:535–539
10. Tanaka T, Fujimoto S, Satoh K, Satoh S, Namanuma K, Midorikawa H (1996) Superselective angiographic findings of ipsilateral middle meningeal artery of chronic subdural hematoma in adults. *Noshinkeigeka* 26:339–347