

The patient underwent spinal cord decompression and C1 lateral mass and C2 pedicle screws and iliac crest bone grafting to correct the instability at the occipitocervical junction (figure, E and F). Direct laryngoscopic examination revealed no tumor recurrence. He received short-term high-dose dexamethasone treatment during the perioperative period, tapered over 1 week. Histopathologic examination of the enhancing nodule and the destroyed anterior arch of C1 showed thickened cortical bone and hypocellular marrow replaced by fibrosis and adipose tissue in multiple foci. The bone was similarly hypocellular, lacking osteoblasts with empty lacunar spaces. There was no evidence of carcinoma or inflammation suggestive of chronic osteomyelitis (figure, I and J).

On his 3-month postoperative follow-up visit, neurologic deficits had entirely resolved apart from 4+ of 5 weakness of left ankle dorsiflexion. Repeat MRI revealed that the medulla oblongata and spinal cord were significantly decompressed, resulting in disappearance of T2 signal abnormality. The C1-odontoid mass was reduced in size without contrast enhancement on T1-weighted images (figure, G and H).

Discussion. The differential diagnosis of an epidural mass in a patient with a history of cancer includes metastasis, radiation-induced secondary tumor, and inflammatory or infectious processes. Osteoradionecrosis (ORN) has not typically been included in this differential diagnosis. Our patient had asymmetrical quadriparesis and an enhancing lesion with mass effect on the spinal cord/medulla. Given the history of oropharyngeal carcinoma and the destructive bony lesion and enhancing mass, metastasis to the skull base and upper cervical vertebrae remained suspect; however, the incidence of spinal metastases from head and neck squamous cell carcinoma is <2%.¹

Another consideration in the differential diagnosis is radiation-induced neoplasm. The most common radiation-induced secondary malignancy is sarcoma, which typically manifests an aggressive pattern of bony destruction.

Radiation-induced myelopathy was also entertained; however, the presence of an epidural mass compressing the spinal cord/medulla and the resolution of spinal cord edema after surgery without spinal cord excluded radionecrosis of spinal cord. Moreover, radiation myelopathy often has enhancement within the cord, which was absent in this case.²

Another diagnostic consideration was ORN. ORN is one of the most serious complications in the treatment of head and neck malignancies and refers to slow-healing, radiation-induced ischemic necrosis of bone associated with soft tissue necrosis of variable extent occurring in the absence of local primary tumor necrosis, recurrence, or metastatic disease.³ The pathophysiology of ORN is believed to be a complex metabolic and homeostatic deficiency

created by radiation-induced cellular injury and fibrosis, characterized by the formation of hypoxic, hypovascular, and hypocellular tissue.⁴ MRI of ORN generally demonstrates T1 hypointensities with variable T2 signal. Unlike tumor, ORN typically does not enhance on postcontrast T1 images. PET may be helpful to differentiate tumor from ORN, which is usually hypometabolic.⁵ There is one case of cervical vertebral ORN reported in the literature; however, it did not cause mass effect on the spinal cord.⁶

Treatment of ORN is difficult. Strategies have included administration of antibiotics, clodronate, and corticosteroids, removal of necrotic bone, hyperbaric oxygen, and combinations of pentoxifylline and tocopherol.⁷ To our knowledge, spinal cord and medullary compression from ORN producing an epidural fibrotic mass has not been previously described. The present report suggests that ORN must be added to the differential diagnosis of enhancing epidural masses originating in a previously irradiated port.

From the Departments of Neurosurgery (Drs. Mut, M. Shaffrey, and C. Shaffrey), Neurology (Dr. Schiff), Pathology (Dr. Miller), and Radiation Oncology (Dr. Larner), University of Virginia, Charlottesville, VA.

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Address correspondence and reprint requests to Dr. Melike Mut, University of Virginia, Department of Neurosurgery, CDW Hospital Drive, P.O. Box 800212, Charlottesville, VA 22903; e-mail: mm2ee@hscmail.mcc.virginia.edu

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References

1. Preciado DA, Sebring LA, Adams GL. Treatment of patients with spinal metastases from head and neck neoplasms. *Arch Otolaryngol Head Neck Surg* 2002;128:539–543.
2. Wang PY, Shen WC, Jan JS. MR imaging in radiation myelopathy. *AJNR Am J Neuroradiol* 1992;13:1049–1058.
3. Wong JK, Wood RE, McLean M. Conservative management of osteoradionecrosis. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997;84:16–21.
4. Marx RE. Osteoradionecrosis: a new concept in its pathophysiology. *J Oral Maxillofac Surg* 1983;41:283–288.
5. Smith WK, Pfeleiderer AG, Millet B. Osteoradionecrosis of the hyoid presenting as a cause of intractable neck pain following radiotherapy and the role of magnetic resonance image scanning to aid diagnosis. *J Laryngol Otol* 2003;117:1003–1005.
6. Lim AA, Karakla DW, Watkins DV. Osteoradionecrosis of the cervical vertebrae and occipital bone: a case report and brief review of the literature. *Am J Otolaryngol* 1999;20:408–411.
7. London SD, Park SS, Gampper TJ, Hoard MA. Hyperbaric oxygen for the management of radionecrosis of bone and cartilage. *Laryngoscope* 1998;108:1291–1296.

Obsessive, prolific artistic output following subarachnoid hemorrhage

M.F.X. Lythgoe, PhD; T.A. Pollak, BA; M. Kalmus, BA; M. de Haan, PhD; and W. Khean Chong, MD

The emergence of artistic skills in the context of brain damage has been documented in patients with frontotemporal dementia (FTD).¹ In this report, we describe an artist whose skills emerged following a sudden-onset brain injury without any associated dementia or impaired verbal abilities. This may represent a window into an alternative mechanism of artistic creativity to that illustrated by patients with FTD.

Case history. A 51-year-old right-handed man was admitted to the hospital (January 2001) with a sudden onset of severe headache. Brain CT on admission showed blood in the anterior parts of the basal cisterns. Angiography demonstrated bilateral middle cerebral artery aneurysms, their irregularity suggested that they were the cause of the hemorrhage. A CT taken 16 days after admission demonstrated no evidence of focal injury.

Our patient worked as a builder and reports that he had a fiery

temper and aggressive personality. At no point was he involved with or interested in the creative arts. Following the subarachnoid hemorrhage he presented with severe depression and complained of a “split-mind disorder.” He also claims to have had difficulty in shaving the left side of his face and his very early drawings show some signs of left hemispatial neglect (figure, A). About 2 weeks after surgery, he began to fill several notebooks with poems and verse (see appendix E-1 on the *Neurology* Web site at www.neurology.org), describing his “split brain,” altered perceptions, and personality. In the following months, he drew hundreds of sketches, mainly of faces, all of them asymmetric (figure, B). This was followed by large-scale drawings on the walls of his house sometimes covering whole rooms (figure, C). He claims the brain injury has left him obsessed with making art and he now spends most of his day painting and sculpting.

Our patient underwent neuropsychological evaluation in May 2004 (table E-1 on the *Neurology* Web site). Verbal and Performance IQ were in the normal range. There were no signs of grossly inappropriate behavior, although he showed a marked verbal disinhibition. Tests of executive function showed some impairments, though not to the extent reported in prior FTD cases. For example, he scored within or above the average range on tests of letter, category, and design fluency; however, his scores dropped dramatically on a more demanding version where he was required to alternate between categories. Generally he showed greatest difficulties in tasks requiring cognitive switching. He was unable to inhibit incorrect responses that either had been previously cor-

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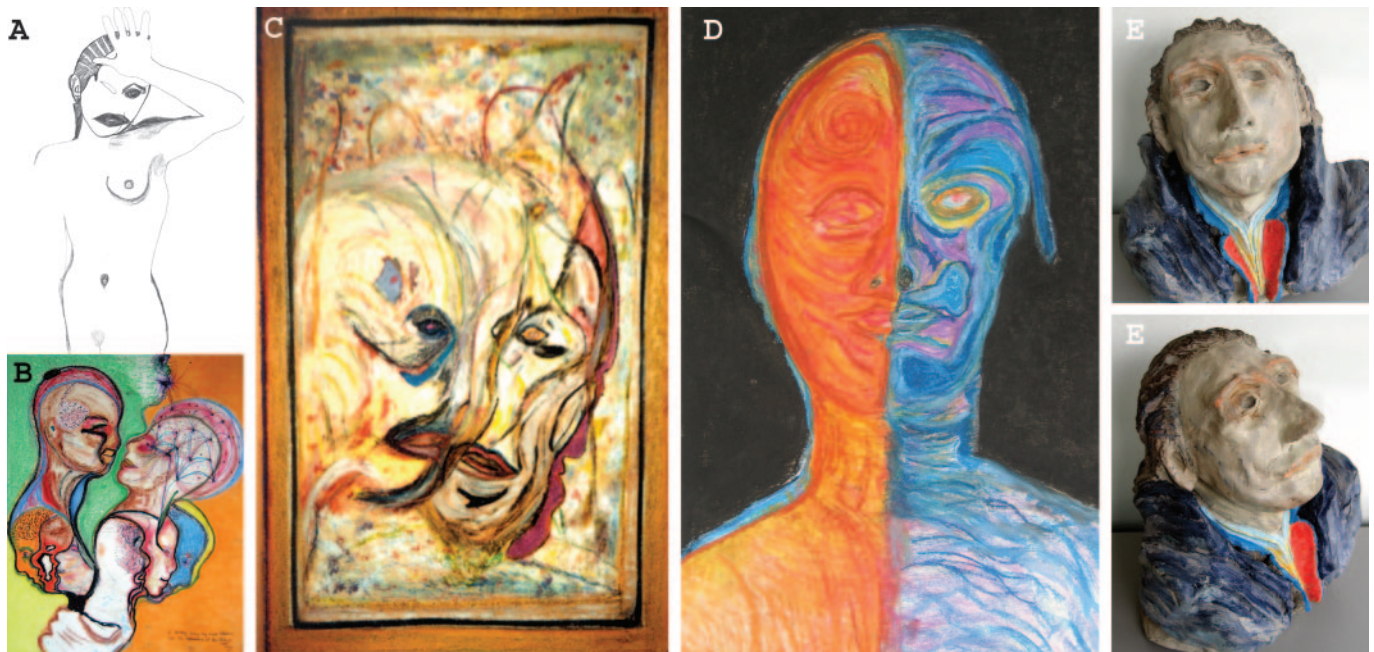


Figure. (A) The artist's first drawings are suggestive of a left hemispatial neglect and would often exclude one half of the subject. (B) His early works are naïve, colorful, and passionate; all of them are asymmetric, describing his split brain in some way. (C) The artist painted this representation of his split brain (including the frame) on the inside wall of his house. He regularly spends 10 hours at a time completing a painting on the wall of his kitchen or living room. (D) Many of his early paintings and poetry (see appendix E-1 on the Neurology Web site at www.neurology.org) comprise of self-investigation of the changes in behavior he had experienced. (E) A recent sculpture created 3 years after his injury demonstrates a striking interpretative skill alongside a growing technical aptitude. In comparison to the earliest drawings this clay head shows a striking congruency, the features are more symmetric, and the colors are more naturalistic.

rect or were semantically associated to previous responses and had a tendency to perseverate. The overall profile is suggestive of mild frontal dysfunction.

Discussion. Some authors have argued that creativity involves a failure in cognitive and even behavioral inhibition.^{2,3} Our patient did show signs of verbal disinhibition, apparent both in his extreme chattiness and his manner of responding during neuropsychological testing. Things he sees and hears seem to trigger a stream of associations which he can find difficult to inhibit. In the context of his artwork, the stream of images and thoughts that are triggered in this way may well be a source of his creativity, at the same time that the process of expression through the artwork may help him to make sense of it.

The emergence or maintenance of visual artistic talent found in some FTD patients has been conceptualized as a "paradoxical functional facilitation"⁴ of the posterior right parietal and temporal cortices (thought to be essential for accurate copying of images and drawing internally imagined images) in the context of an impairment in "language dominant patterns of thinking organized in the dominant frontal and anterior temporal regions."⁵ It is possible that whatever frontal damage our patient sustained led to a relative disinhibition or paradoxical functional facilitation of these areas.

However, our patient had normal Verbal IQ, he remained very conversational, and has written hundreds of poems, suggesting that left anterior-temporal function has been preserved. Whatever the damage sustained, this case would appear to demonstrate that paradoxical functional facilitation of artistic talent can occur in the context of relative dysfunction of areas other than dominant areas specialized for language. Further, our patient showed some left hemispatial neglect, which is suggestive of dysfunctional right frontal/parietal connection.⁶

It may be that perseverative tendencies are central to understanding⁷ the character of our patient's creative drive and could be integral to his sustained and voluminous output of thematically similar work. The emergence of artistic skills following subarachnoid hemorrhage may represent another platform into the mecha-

nisms of artistic creativity and an unrecognized attribute of this type of brain injury.

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From the RCS Unit of Biophysics (Dr. Lythgoe, and T.A. Pollak and M. Kalmus) and Developmental Cognitive Neuroscience (Dr. de Haan), Institute of Child Health; and Department of Radiology (Dr. Khean Chong), Great Ormond Street Hospital, London, UK.

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Address correspondence and reprint requests to Dr. Mark F. Lythgoe, RCS Unit of Biophysics, Institute of Child Health, University College London, London, WC1N 1EH UK; e-mail: mlythgoe@ich.ucl.ac.uk

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References

1. Miller BL, Cummings JL, Mishkin F, et al. Emergence of artistic talent in frontotemporal dementia. *Neurology* 1998;51:978–981.
2. Eysenck H. *Genius: the natural history of creativity*. Cambridge: Cambridge University Press, 1995.
3. Carson SH, Peterson JB, Higgins DM. Decreased latent inhibition is associated with increased creative achievement in high-functioning individuals. *J Pers Soc Psychol* 2003;85:499–506.
4. Kapur N. Paradoxical functional facilitation in brain-behaviour research. A critical review. *Brain* 1996;119:1775–1790.
5. Mell JC, Howard SM, Miller BL. Art and the brain. The influence of frontotemporal dementia on an accomplished artist. *Neurology* 2003;60:1707–1710.
6. Doricchi F, Tomaiuolo F. The anatomy of neglect without hemianopia: a key role for parietal-frontal disconnection? *Neuroreport* 2003;14:2239–2243.
7. Cybulska E. Bolero unravelled. A case of musical perseveration. *Psychiatr Bull* 1997;21:576–577.