

Case report

Bilingual aphasia due to spontaneous acute subdural haematoma from a ruptured intracranial infectious aneurysm

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Received 10 December 2007; received in revised form 10 April 2008; accepted 8 May 2008

Abstract

We report a case of spontaneous subdural haematoma due to ruptured intracranial infectious aneurysm, presenting with bilingual aphasia and illustrating differential language recovery. A 62-year-old right-handed bilingual gentleman, with a diagnosis of infective endocarditis, developed headache and became expressively aphasic in the English language. Three days later he was receptively and expressively aphasic in both English and Arabic. Cranial MRI scans showed a left-sided acute subdural haematoma with mass effect and midline shift. Contrast CT brain scans showed an enhancing speck adjacent to the clot and cerebral angiogram confirmed a distal middle cerebral artery aneurysm. He underwent image-guided craniotomy, evacuation of the subdural haematoma and excision of the aneurysm. Histopathological examination was consistent with an infectious intracranial aneurysm. Postoperatively his aphasia did not improve immediately. He had widened pulse pressure due to severe aortic regurgitation, confirmed on echocardiography. He underwent aortic valve replacement and mitral valve repair, following which his aphasia recovered gradually. Initially the recovery of his language was limited to Arabic. About a week later he recovered his English language as well. At 3-year follow-up he is doing well and has no neurological deficits. His aphasia has recovered completely. The present case is unique because of (a) presence of pure subdural haematoma, and (b) the differential susceptibility and recovery of native (L1) and acquired language (L2) in presence of a common pathology. The neurology of language in a bilingual is analysed and possible mechanisms discussed.

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Keywords: Bilingual aphasia; Infectious intracranial aneurysm; Mycotic aneurysm; Subdural haematoma

1. Introduction

A ruptured intracranial infectious aneurysm (IIA) usually produces subarachnoid haemorrhage (SAH) or intracerebral haemorrhage (ICH). There have been very few reports of IIA producing subdural haemorrhage (SDH) associated with ICH and only a single report of pure SDH due to ruptured IIA [1–6]. We report the second case of spontaneous SDH due to ruptured IIA. Our patient presented with bilingual aphasia with early involvement and delayed recovery of the second acquired language (L2) compared to his primary/native language (L1). Several investigators have studied the cortical

representation of different languages in multilingual people. Although no consensus has been reached, it is generally believed that the cortical representation of L1 is different from that of L2, especially if L2 is acquired after age seven [7]. Aphasia specific to one language in a bilingual patient has been described following various neurological insults, which could impair L1 or L2 cortex. The uniqueness of the present report is the differential involvement and recovery of L1 and L2 in presence of a common neurological insult.

2. Case report

A 62-year-old right-handed late bilingual Egyptian gentleman was investigated extensively in Egypt for pyrexia of unknown origin of 6 months duration. Having failed to come to a diagnosis, he was referred to physicians in the UK for

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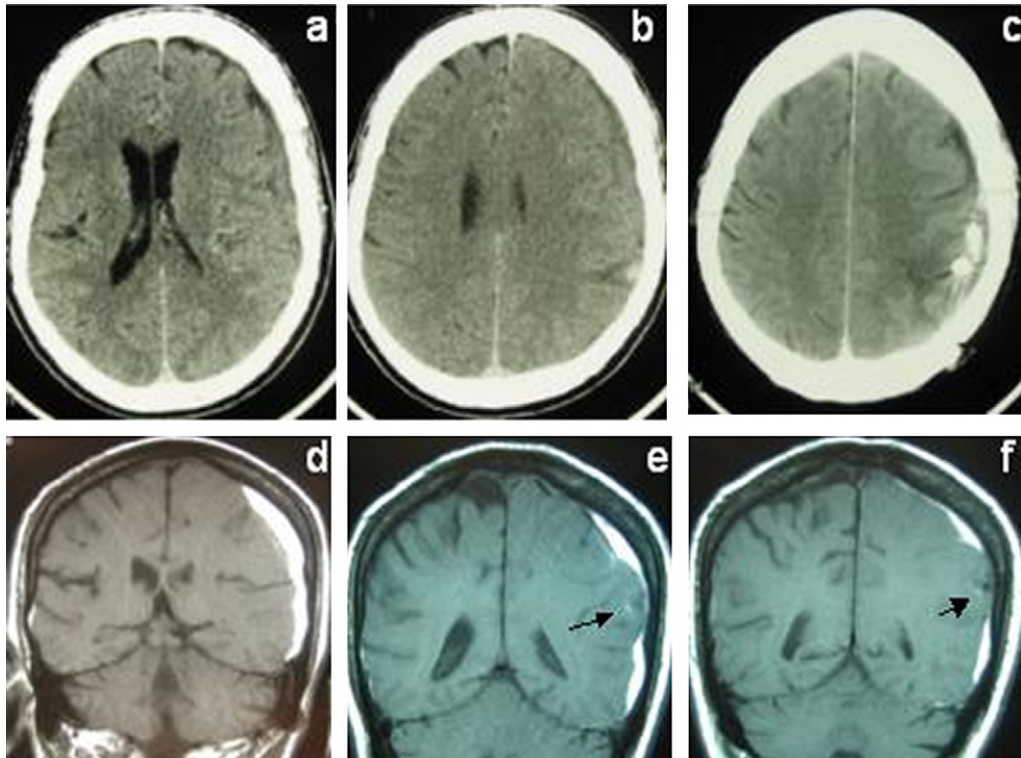


Fig. 1. (a and b) Preoperative CT scan brain showing the left frontoparietal subdural haematoma, (c) postoperative CT scan revealing the aneurysm clips and very minimal residual subdural haematoma and (d, e and f) preoperative MR scan revealing the subdural haematoma in left frontoparietal region. Notice the flow void (black arrows) in (e) and (f) adjacent to the posterior margin of the acute subdural haematoma caused by the ruptured aneurysm, as well as the absence of any associated intracerebral haematoma.

further investigation and management. He had previously developed an aneurysm arising from the left distal radial artery, which was excised a few months prior to arrival in the UK. At a local hospital in Winchester, a diagnosis of infective endocarditis (IE) was made and he was started on benzyl penicillin 2 mega units 4 hourly and gentamycin 80 mg 12 hourly. Blood culture had grown *Streptococcus sanguinis* and echocardiogram revealed aortic valve vegetations. During his hospital stay he developed headaches and later some slurring of speech. On examination, his blood pressure was 140/50 mm Hg with clinical evidence of aortic regurgitation. Neurologically, he was conscious, alert and orientated. His English and Arabic speech were initially reported as appearing “slurred” but he was able to follow simple commands. He did not have any other neurological deficits. With the exception of slurring, his verbal production and comprehension in his native Arabic language were initially preserved. MRI scan of the brain revealed an acute subdural haematoma (SDH) in the left frontoparietal region with mass effect as well as a suspicion of a flow void at the posterior margin of the SDH (Fig. 1d, e and f). CT scan with contrast revealed the subdural haematoma with an enhancing lesion adjacent to the SDH (Fig. 1a and b). At the Wessex Neurological Centre, where he was transferred for further management, he underwent cerebral angiography, which showed an aneurysm arising from the left distal middle cerebral artery (MCA), M4

segment, at the posterior margin of the SDH (Fig. 2). His neurological condition deteriorated. He initially had word finding difficulty in English with normal Arabic. His speech deteriorated rapidly over 48 h, initially in English and later in Arabic, so that he could neither speak nor comprehend either language. On bedside examination his other cognitive functions appeared grossly intact. He underwent an image-guided craniotomy, evacuation of the acute SDH and excision of the aneurysm. At surgery the aneurysm was adherent to the dura. The neck of the aneurysm involved the bifurcation of the distal M4 and was very friable; hence clipping of the aneurysm neck to preserve the parent vessel was abandoned in favour of complete excision of the aneurysm. Histological examination was consistent with that of an infectious intracranial aneurysm (IIA). Postoperatively he continued to have aphasia in both languages.

He was transferred to the cardiology team for urgent management of the endocarditis and aortic valve incompetence. He underwent aortic valve replacement and mitral valve repair with excision of the vegetations 3 days after the craniotomy. Following the repair, as the wide pulse pressure was normalised and his mean arterial pressure rose, his aphasia began to improve. Initially his speech was non-fluent in Arabic and his communication was restricted to nodding and saying “yes”. His English language was globally impaired. His Arabic rapidly recovered to a more typically fluent

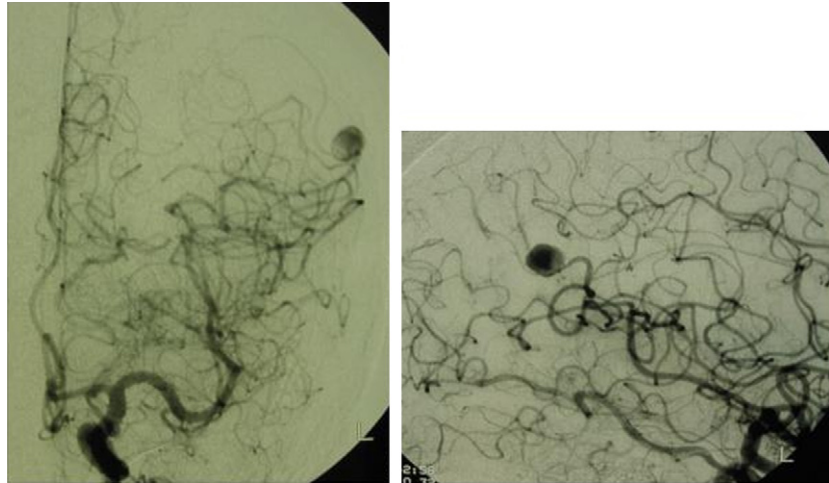


Fig. 2. Preoperative left carotid angiogram demonstrating the IIA in the left distal middle cerebral artery.

aphasic profile. His family initially described him as being talkative but making little sense and using words that were unrelated to the context of a conversation. His comprehension was poor. However, after 3 days he was communicating effectively in Arabic and was able to participate in conversations. His English language remained globally impaired throughout this time. He had grave expressive difficulties in English and his communication was restricted to nodding and saying “yes” with poor comprehension of commands and questions. His communication with the ward staff was dependent on the assistance of a translator. About a week later his English began to recover and he was observed to follow English speech on the radio and to be using words appropriately. Three weeks after his cardiac surgery no residual language deficits were apparent in English or Arabic. Repeat CT scan of the brain showed a residual thin SDH without any mass effect (Fig. 1c). Check cerebral angiogram 4 weeks after the initial one, confirmed complete excision with no new aneurysm seen elsewhere (Fig. 3). At 3-year follow up the patient is doing well and has no neurological deficits. His speech disturbance has recovered completely.

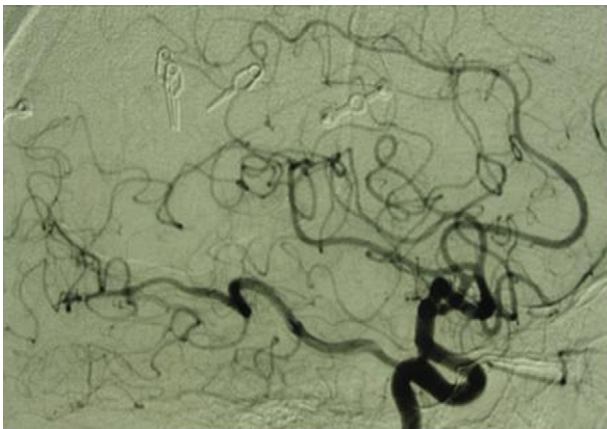


Fig. 3. Postoperative angiogram showing the aneurysm clips in situ. The aneurysm has been excluded from circulation and no new aneurysm seen.

3. Discussion

This report is unique because of (a) the occurrence of pure acute SDH due to ruptured IIA and (b) the differential involvement and recovery of L1 and L2.

The incidence of acute SDH related to rupture of IIA or berry aneurysms is very low. IIA accounts for only 2.5–4.5% of all intracranial aneurysms [4]. Despite its peripheral location, only few cases of SDH caused by ruptured IIA have been reported (Table 1). We could find only one similar case of pure SDH due to ruptured IIA, whereas others had ICH as well, in addition to SDH [1]. However in that case the diagnosis was based on angiographic and operative findings. CT or MR scans were not done. There have been five other reports of acute SDH due to ruptured IIA, all of whom had an associated ICH (Table 1) [2–6].

The presence of adhesions between the aneurysm and the arachnoid membrane may allow the aneurysm to bleed into the subdural space. Blood from a ruptured aneurysm can also disrupt the arachnoid layer and force its way into the subdural space [5,6]. In the present case the aneurysm, was indeed adherent to the dura, probably resulting from

Table 1
Ruptured IIA producing SDH or SDH with ICH

Author	Year	Type of haemorrhage	Location of IIA
King [1]	1960	SDH only	Left angular artery
Bamford et al. [2]	1986	ICH and SDH	Right temporooccipital artery
Bandoh et al. [3]	1987	ICH and SDH	Distal right MCA
Barami and Ko [4]	1994	ICH and SDH	Left parietooccipital branch of PCA
Piastra et al. [5]	2000	ICH and SDH	Distal left MCA
Yamakawa et al. [6]	2003	ICH and SDH	Distal left MCA
Present case	2007	SDH only	Distal left MCA

IIA, infectious intracranial aneurysm; SDH, subdural haematoma; ICH, intracerebral haematoma; MCA, middle cerebral artery; PCA, posterior cerebral artery.

Table 2
Cortical representation of L1 and L2

Author/reference	Year	Investigation	Task to stimulate speech area	Cortical representation of L1 and L2
Ojemann and Whitaker [8]	1978	ESM	Intraoperative cortical stimulation	Both overlapping and distinct areas of the cortex
Klein et al. [9]	1994	PET scan	Word processing	No difference in the activation pattern
Perani et al. [10]	1996	PETscan	Story listening	Distinct cortical areas
Dehaene et al. [11]	1997	fMRI	Listening to stories	Dissociation between cortical areas
Kim et al. [12]	1997	fMRI	Silent internally expressive linguistic task	Broca-Separate in late bilinguals and common in early bilinguals Wernicke-Common in both early and late bilinguals
Chee et al. [13]	1999	fMRI	Cued word generation task	Common cortical representation
Klein et al. [14]	1999	PET scan	Verb generation	Common areas of activation
Illes et al. [15]	1999	fMRI	Semantic judgement task	Similar cortical areas
Pouratian et al. [16]	2000	iOIS	Visual object naming	Distinct cortical areas
Hernandez et al. [17]	2000	fMRI	Picture naming task	Similar cortical areas
Pu et al. [18]	2001	Event related fMRI	Verb generation	Shared mechanism
Simos et al. [19]	2001	Magnetic source imaging	Auditory and visual comprehension	Distinct areas
Roux and Tremoulet [20]	2002	ESM	Reading, counting and word-retrieval	Both common and language-specific areas
Lucas et al. [21]	2004	ESM	Confrontation object naming paradigm	Distinct and shared language specific sites. L2 areas mainly in posterior temporal and parietal lobe
Walker et al. [22]	2004	ESM	Object naming task	Both common and distinct areas
Bello et al. [23]	2006	ESM	Counting and oral naming tasks	Distinct cortical areas

ESM, electrical stimulation mapping; PET, positron emission tomography; fMRI, functional magnetic resonance imaging; iOIS, intraoperative optical imaging of intrinsic signals.

infection or a prior subclinical bleed. This allowed the aneurysm to rupture into the subdural space without any subarachnoid or parenchymal bleed.

In the present case, the IIA developed in presence of active bacterial endocarditis. Although his symptoms of endocarditis were of 6 months duration, the diagnosis was missed and hence adequate treatment was not instituted. An IIA rupture usually occurs during the course of endocarditis. Rupture weeks or months after adequate treatment is unusual, although there are reports where IIA ruptured 6 months after cure of bacterial endocarditis [6]. Interestingly our case had in addition an aneurysm arising from distal radial artery that was excised a few months prior to the onset of the neurological illness.

The present case also illustrates the differential vulnerability and recovery of L1 and L2 in presence of a common neurological insult. The left sided acute SDH exerted a mass effect on the underlying cortex. This was compounded by the reduced cerebral perfusion due to a low mean arterial pressure from the severe aortic regurgitation. It was only after surgical correction of the aortic regurgitation that the mean arterial pressure stabilised, resulting in rapid recovery of language dysfunction. There was no evidence of vasospasm on the cerebral angiography, or of any ischaemic areas on the preoperative CT brain scan.

Although the cortical representation of language in a monolingual is reasonably clear, the neural substrates of bilingualism are largely unknown. Cortical representation of language in multilinguals has been the topic of intense ongoing research [8–23]. Language recovery patterns following

aphasia in bilinguals have been well documented since the late 19th century. Pitre's rule (initial recovery of the most intensively used language) and Ribot's law (recovery of the earliest acquired or native language) characterise the differential recovery of L1 and L2 [24]. Paradis [24] has identified various recovery patterns in bilingual aphasics. L1 and L2 can be affected in parallel, differentially or selectively. The findings in our patient were in accordance with both Pitre's rule and Ribot's laws with the most intensively used and earliest acquired language (Arabic) being affected later and recovering first.

Both similar and distinct cortical areas have been reported to subserve language functions in a multilingual. Klein et al. [9], Chee et al. [13], Illes et al. [15], and Hernandez et al. [17] have argued that their representation in cortex is identical (Table 2). However, many other studies based on clinical cases, electrical stimulation mapping, optical intrinsic signal imaging, functional MRI, and PET studies suggest that L1 and L2 implicate distinct cortical areas (Table 2) [8,10–12,14,16,18–23]. It has been argued that the variability in the findings depends upon the age of acquisition of L2, proficiency of L2, investigative methodology and the task used to activate the speech area (Table 2).

Several possible mechanisms could explain the differential language involvement in our patient. Dehaene et al. [11] proposed that the right hemisphere may initially be dominant for L2. As one becomes more proficient in a language, the left hemisphere may become more involved [11]. This explanation seems somewhat unlikely in our case, for whom the pathology was restricted to the left hemisphere.

The other possible reason could be sparing of subcortical structures, which are essential for L1, and greater involvement of neocortical structures essential for L2 [25]. However the parallel pattern of recovery that we documented in L1 and L2 suggests a common neural substrate.

In the present case, the pattern of language recovery would be most consistent with a common distributed representation supporting both L1 and L2 with a relatively more robust neuronal circuitry for L1 [8,11,26]. Damage to a distributed system exhibits “graceful degradation” with less frequently activated representations being more susceptible to the effects of damage anywhere within the distributed network [26]. In the case of a global aphasia recovering to a fluent aphasia with inappropriate word choice it seems most likely that temporal lobe language areas bore the brunt of our patient’s neurological dysfunction. As the distributed language system recovered, “graceful recovery” was exhibited and an ability to speak was followed by recovery of comprehension and then by normal speech with L1 being restored before L2.

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