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Aphasia Owing to Subcortical Brain Infarcts in Childhood

Ariel Gout, MD, PhD; Nathalie Seibel, BA; Constance Rouvière, BA; Béatrice Husson, MD; Brigitte Hermans, BA; Nicole Laporte, BA; Hazim Kadhim, MD, PhD; Cécile Grandin, MD, PhD; Pierre Landrieu, MD; Guillaume Sébire, MD, PhD

ABSTRACT

The aim of this study was to further define the clinical features of subcortical aphasia in children with deep brain infarcts and to define the sequelae associated with childhood strokes. We retrospectively studied nine children with left subcortical brain infarcts who presented with acquired language disorder and underwent language investigations based on standardized tests. Stroke in these patients involved the left internal capsule, lenticular or thalamic nuclei, or a combination of these. Early aphasic manifestations following the deep cerebral infarcts affected language expression. These included mutism, nonfluent speech, word finding difficulties, and phonemic and semantic paraphasia. Speech comprehension was generally more preserved. All patients subsequently improved, although variably; sequelae such as dysfluency, word finding difficulties, and written language learning impairment could be detected through standardized tests in six of them (all younger than 6 years at the time of the infarct). Two of the three remaining patients (both older than 6 years at the time of the infarct) had a full recovery. Our study confirms the concept of childhood subcortical aphasia, depicts the linguistic profile in these patients, and sustains the indication of systematic formal language assessment during the follow-up of all children with subcortical infarct involving the dominant hemisphere. (*J Child Neurol* 2005;20:1003–1008).

Most of the studies about aphasic children include rather heterogeneous conditions with regard to the etiology of underlying aphasia; these comprise cranial trauma, cerebral tumors, encephalitis, and ischemic and hemorrhagic stroke.^{1–7} The diffuse nature of the underlying lesions prevailing in the majority of these pathologic

states and the lack of accurate radiologic investigations, namely magnetic resonance imaging (MRI) in most of the studies, made precise anatomoclinical correlation difficult. So, the profile of language impairments owing to small and anatomically well-defined acquired brain lesions, such as subcortical stroke, is still poorly described in childhood. Brain infarcts, relatively rare in children—an incidence of 8 per 100,000 children⁸—often affect basal ganglia and adjacent white matter. Featuring the cognitive outcome of such lesions is of importance when considering the benefit versus risk and cost balances for therapeutic interventions in childhood stroke, such as immediate anticoagulation or early rehabilitation. Moreover, subcortical infarcts in childhood offer the opportunity to explore the role of deep brain structures in the development of language. In this study, we looked at neuropsychologic and radiologic findings (1) to portray the linguistic profile of subcortical aphasia in children and compare it with the profile of adults, (2) to define the potential burden of language sequelae generated by ischemic strokes in childhood, and (3) to explore the role of subcortical structures, particularly the basal ganglia and internal capsule, in the processing of speech.

PATIENTS AND METHODS

Sixteen children were admitted consecutively between 1985 and 1996 in a pediatric stroke referral center (CHU Bicêtre, France) for ischemic infarcts

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From the Service de Neurologie (Drs Gout, Landrieu, and Sébire and Mrs Seibel and Rouvière), Département de Pédiatrie, CHU Bicêtre, Paris, France; Service de Radiologie (Dr Husson), Département de Pédiatrie, CHU Bicêtre, Paris, France; Unité de Neurologie du Développement (Mrs Hermans and Laporte and Drs Kadhim and Sébire), Université Catholique de Louvain, Brussels, Belgium; Unité de Neuropathologie (Dr Kadhim), Service d'Anatomopathologie, CHU Brugmann-HUDERF, Université Libre de Bruxelles, Brussels, Belgium; Service de Neuroradiologie (Dr Grandin), Cliniques Universitaires Saint Luc, Université Catholique de Louvain, Brussels, Belgium; Service de Neurologie (Dr Sébire), Département de Pédiatrie, Université de Sherbrooke, Sherbrooke, QC, Canada.

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Address correspondence to Dr Guillaume Sébire, Université de Sherbrooke, 12e avenue Nord, Sherbrooke, QC J1H 5N4, Canada. Tel: 819-346-1110; fax: 819-564-5398; e-mail: Guillaume.Sebire@usherbrooke.ca.

involving the basal ganglia, thalamus, or adjacent white matter. These patients met the following criteria for infarction: prolonged neurologic deficit with sudden onset and an MRI and/or a computed tomographic (CT) scan showing brain lesions in an arterial territory. Eleven patients had left-hemisphere lesions, and the remaining five had right lesions. Nine of the eleven patients with a left-hemisphere lesion initially presented with aphasic symptoms, whereas none of the five patients with a right lesion did. We retrospectively studied the nine aphasic patients. None of them had a neurologic history, their psychomotor development had been normal, their native language was French, and they were all right-handed (except patient 7, who was too young). All children underwent cerebral imaging, namely CT ($n = 8$) and/or MRI ($n = 5$; axial, coronal, and sagittal sections, T_1 - and T_2 -weighted sequences). Conventional arteriography performed in eight patients (all except patient 6) within a few days after stroke disclosed lesions involving the distal part of the internal carotid artery, the initial segment of the middle cerebral artery, or the proximal segments of insular or temporal branches of the middle cerebral artery. According to the clinical history and to the angiographic aspect, blood flow disruptions were related to intracranial carotid dissection (patients 1 and 7), post-varicella-zoster virus acute vasculitis (patients 2, 5, and 9), emboli from cardiac origin (patients 3 and 6), or stenosis of the middle cerebral artery (patient 4). The overall follow-up ranged from 3 months to 7 years (mean 2 years) for language assessment and to 12 years for schooling.

All patients underwent full language investigations according to their age. The speech therapist proposed the *Epreuve pour l'évaluation du langage*, the *Test de vocabulaire actif et passif*, the Token Test for Children, the Clinical Evaluation of Language Fundamentals (CELF) test, the Borel Maisony written language test, and the Echelle d'évaluation de l'aphasie (BDAE) and the 15 mots de Rey verbal memory tests.⁹⁻¹⁴ Children suspected of presenting with global cognitive dysfunction (patients 1, 6, and 9) were subjected to an intellectual evaluation (Wechsler Intelligence Scale for Children [WISC]: Wechsler Preschool and Primary Scale of Intelligence [WPPSI], WISC-R, or WISC-III).¹⁵

RESULTS

Clinical Features

The nine aphasic patients (mean age 6.5 years; median age 5.5 years; range 2–14 years; male to female ratio 1:0.8) presented with varying degrees of right motor deficits. Patients 4, 6, 8, and 9 had hemiplegia. Patients 2, 3, and 7 had hemiparesis. Patient 5 presented with an isolated facial paralysis, and patient 1 presented with facial paralysis and right hemineglect. Patient 4 also had a right lateral homonymous hemianopsia. There was complete regression of the motor deficits after a few days (patients 1, 2, and 8) or a few months (patients 4 and 5). Four patients showed motor sequelae with the persistence of arm dystonia (patients 3, 6, 7, and 9). There was no recurrence of stroke. All patients were treated with antiplatelet agents.

Anatomic Localization of the Cerebral Infarcts

Three patients presented with left lesions involving the head of the caudate nucleus, the putamen, the anterior limb of the internal capsule (patient 5), and the pallidum (patients 3 and 7). Two patients presented with more posterior lesions located in the posterior limb of the internal capsule (patients 2 and 8) and in the thalamus (patient 8). Four patients displayed lesions involving both the anterior and the posterior regions of the basal nuclei (patients 1, 4, 6, and 9) (Table 1 and Figure 1).

Language Anomalies Within the First Month

The language anomalies observed in these patients shared many common features, namely, reduction in spontaneous speech, word finding difficulties, paraphasia, and written language anomalies. In the acute phase, mutism was noted (patients 2, 6, and 7), lasting from 2 to 12 days, as well as reduced spontaneous speech (patients 1, 3, and 9). Dysarthria or slurred speech was observed in three children (patients 4, 5, and 6). At the first formal language assessment, seven patients showed a deficit in oral expression and nonfluent speech, associated with word finding difficulties (designation was far better than naming in patients 1, 3, 4, 6, 7, and 9). Semantic fluency investigated immediately after the cerebral infarct in four patients was deficient for all of the semantic categories tested (animals, clothes, and food; patients 1, 3, and 9) and for one of the categories tested (clothes; patient 8). Positive aphasic signs were frequently observed: semantic paraphasia, phonemic paraphasia, and neologisms (patients 1, 3, 5, 6, 8, and 9). Phonologic disorders (omissions and simplifications) were rare and transient (patients 2 and 5). No major syntactic difficulties were detected. Speech comprehension difficulties were detected in four patients. In three children (patients 1, 3, and 8), understanding of everyday conversation was normal, whereas deficits were highlighted within formal tests. Patient 4 presented with a profile of sensorial aphasia: he had serious and global deficit in oral comprehension, was unable to repeat, and had an auditory comprehension deficit. Disorders of written language (both spelling and reading) were seen immediately after the infarct in the three school-aged patients: patient 4 could read vowels only, patient 3 had poor reading comprehension, and patient 1 presented with a spelling and reading disorder.

Outcome

In six of the nine patients studied, sequelae developed (see Table 1). Two outcome patterns were observed in five patients of preschool age at the time of their infarct. In three of them, speech disorders had been detected since the initial period. These deficits later improved, but lexical access problems (patient 6) or reduced lexical stock (patients 5 and 8) persisted. In two other preschool children (patients 2 and 9), language difficulties were secondarily noticed at the time of the acquisition of new skills. Thus, in patient 2, although the initial formal assessment of speech completed 2 years after the stroke had been quite normal, later difficulties appeared: acquisition of written language was delayed, and by the age of 8 years (2 years after written language learning; 5 years after stroke), he was still unable to read and had global school difficulties. In patient 9, whereas word naming difficulties were no longer noticeable 18 months after stroke, paraphasia reappeared 5 years later. Semantic fluency, although improved over 5 years, still remained deficient, and a spelling and reading disorder persisted. In patient 7 (2 years old at the time of the infarct), the relatively short follow-up (18 months) would not enable us to definitely conclude about his apparent recovery and to say anything about his abilities to learn written language.

Among the three school-aged children at the time of their infarct, patient 4 recovered well, patient 3 had a follow-up too short (3 months) to conclude, and patient 1 (8 years old) developed sequelae; although his speech returned to normal within 12 months, he continued to suffer from a spelling disorder.

Table 1. Main Radiologic and Linguistic Features

Patient	Age (yr, mo)	Topography of Brain Lesions*						Follow-up	Evolution of Language Anomalies
		Put	Pal	CN	ICA	Th	ICP	SOC	
1	8, 3	—	+	—	—	—	+	+	D6 Anomia in conversation and elaborate language only; oral comprehension deficit in Token Test; difficulties in spelling and narrative writing, verbal memory for words subnormal; PIQ = 88, VIQ = 85 Repetition disorders for phonologically complex words, poor language elaboration (definition), spelling disorders; attention disorder; stay down at school
2	2, 9	—	—	—	—	—	+	—	M3 M12 D1 M24 M48 M60 D10 M3 M24 D11 M11 M3 Y7 D1 M48 D12 D19 M12 Y3 D1 M1 M4 M18 D5 M7 Y3 D11 M3 M18 Y5 D1 M3 M18 Y5
3	14, 3	+	+	+	+	—	—	—	Mutism 48 h, simple comprehension normal; nonfluent aphasia Mild difficulties in naming, repetition, and syntax; comprehension and verbal retention normal Mild phonologic disorders; naming, designation, elaborate language and fluency reduced; no written language and reading after 1 yr of elementary school Deficient reading and global difficulties at school, attention deficit Hypospontaneity, hypoinformativity, reduced fluency, anomia, semantic paraphasia, deficit elaborate language (definition, sentences generating); verbal retention deficit (for sentences and digit), disturbed oral comprehension tests; poor reading comprehension; nonfluent aphasia Category naming difficulties (body part); reduced phonemic fluency, poor verbal retention for sentences and story Apprenticeship orientation Anomia, reading difficulties (recognizes vowels only) Mild dysarthria; repetition disorders (for syllables, digits, and nonwords) with auditory comprehension impairment; reduced digit retention; sensorial aphasia Normal except auditory comprehension for long or complex stimuli Normal except nonsemantic items retention (digit span: 4/20); retention of auditory story normal Dysarthria, deficient elaborate language (sentences generation); mild, nonfluent aphasia Normal except elaborate language: poor vocabulary in definition, dysyntaxia, and hypoinformativity in narration; correct naming and fluency tests Mutism for 12 d Slurred and very reduced speech; anomia, phonemic and semantic paraphasia; VIQ = 75, PIQ = 98 Persistent anomia and phonemic paraphasia; poor narrative capacity; written language is acquired Specialized heart disease school Mutism Loss of previous language abilities; says a few isolated words; nonfluent aphasia Severely reduced and agrammatic language, phonologic disorders; normal repetition Normal; some persistent phonologic simplifications Fluent, noninformed speech; naming difficulties, phonemic and semantic paraphasia, phonologic deficit, syntactic errors; repetition and oral comprehension impairment; fluent aphasia Hypoinformativity, paraphasia, phonologic deficit, repetition deficit, complex comprehension impairment; poor vocabulary Hypoinformativity, poor vocabulary, impaired elaborate language (inconsistent definitions); written language is acquired; normal academic achievement Mutism Reduced spontaneous speech, anomia, paraphasia; nonfluent aphasia Fluent and informative speech; mild anomia in naming and fluency tests; normal narrative capacity Normal oral speech but reading and spelling difficulties Word finding difficulties in spontaneous speech, paraphasia and perseverations in naming and fluency tests; reading and spelling deficits; low verbal retention; VIQ = 109, PIQ = 85, GIQ = 97; 4th yr of elementary school (stayed down in the 1st yr)
4	10, 6	+	—	—	—	—	+	+	Mutism Anomia, reading difficulties (recognizes vowels only) Mild dysarthria; repetition disorders (for syllables, digits, and nonwords) with auditory comprehension impairment; reduced digit retention; sensorial aphasia Normal except auditory comprehension for long or complex stimuli Normal except nonsemantic items retention (digit span: 4/20); retention of auditory story normal Dysarthria, deficient elaborate language (sentences generation); mild, nonfluent aphasia Normal except elaborate language: poor vocabulary in definition, dysyntaxia, and hypoinformativity in narration; correct naming and fluency tests Mutism for 12 d Slurred and very reduced speech; anomia, phonemic and semantic paraphasia; VIQ = 75, PIQ = 98 Persistent anomia and phonemic paraphasia; poor narrative capacity; written language is acquired Specialized heart disease school Mutism Loss of previous language abilities; says a few isolated words; nonfluent aphasia Severely reduced and agrammatic language, phonologic disorders; normal repetition Normal; some persistent phonologic simplifications Fluent, noninformed speech; naming difficulties, phonemic and semantic paraphasia, phonologic deficit, syntactic errors; repetition and oral comprehension impairment; fluent aphasia Hypoinformativity, paraphasia, phonologic deficit, repetition deficit, complex comprehension impairment; poor vocabulary Hypoinformativity, poor vocabulary, impaired elaborate language (inconsistent definitions); written language is acquired; normal academic achievement Mutism Reduced spontaneous speech, anomia, paraphasia; nonfluent aphasia Fluent and informative speech; mild anomia in naming and fluency tests; normal narrative capacity Normal oral speech but reading and spelling difficulties Word finding difficulties in spontaneous speech, paraphasia and perseverations in naming and fluency tests; reading and spelling deficits; low verbal retention; VIQ = 109, PIQ = 85, GIQ = 97; 4th yr of elementary school (stayed down in the 1st yr)
5	5, 6	+	—	—	+	—	—	—	Mutism Anomia, reading difficulties (recognizes vowels only) Mild dysarthria; repetition disorders (for syllables, digits, and nonwords) with auditory comprehension impairment; reduced digit retention; sensorial aphasia Normal except auditory comprehension for long or complex stimuli Normal except nonsemantic items retention (digit span: 4/20); retention of auditory story normal Dysarthria, deficient elaborate language (sentences generation); mild, nonfluent aphasia Normal except elaborate language: poor vocabulary in definition, dysyntaxia, and hypoinformativity in narration; correct naming and fluency tests Mutism for 12 d Slurred and very reduced speech; anomia, phonemic and semantic paraphasia; VIQ = 75, PIQ = 98 Persistent anomia and phonemic paraphasia; poor narrative capacity; written language is acquired Specialized heart disease school Mutism Loss of previous language abilities; says a few isolated words; nonfluent aphasia Severely reduced and agrammatic language, phonologic disorders; normal repetition Normal; some persistent phonologic simplifications Fluent, noninformed speech; naming difficulties, phonemic and semantic paraphasia, phonologic deficit, syntactic errors; repetition and oral comprehension impairment; fluent aphasia Hypoinformativity, paraphasia, phonologic deficit, repetition deficit, complex comprehension impairment; poor vocabulary Hypoinformativity, poor vocabulary, impaired elaborate language (inconsistent definitions); written language is acquired; normal academic achievement Mutism Reduced spontaneous speech, anomia, paraphasia; nonfluent aphasia Fluent and informative speech; mild anomia in naming and fluency tests; normal narrative capacity Normal oral speech but reading and spelling difficulties Word finding difficulties in spontaneous speech, paraphasia and perseverations in naming and fluency tests; reading and spelling deficits; low verbal retention; VIQ = 109, PIQ = 85, GIQ = 97; 4th yr of elementary school (stayed down in the 1st yr)
6	5, 4	+	+	—	—	—	+	—	Mutism Anomia, reading difficulties (recognizes vowels only) Mild dysarthria; repetition disorders (for syllables, digits, and nonwords) with auditory comprehension impairment; reduced digit retention; sensorial aphasia Normal except auditory comprehension for long or complex stimuli Normal except nonsemantic items retention (digit span: 4/20); retention of auditory story normal Dysarthria, deficient elaborate language (sentences generation); mild, nonfluent aphasia Normal except elaborate language: poor vocabulary in definition, dysyntaxia, and hypoinformativity in narration; correct naming and fluency tests Mutism for 12 d Slurred and very reduced speech; anomia, phonemic and semantic paraphasia; VIQ = 75, PIQ = 98 Persistent anomia and phonemic paraphasia; poor narrative capacity; written language is acquired Specialized heart disease school Mutism Loss of previous language abilities; says a few isolated words; nonfluent aphasia Severely reduced and agrammatic language, phonologic disorders; normal repetition Normal; some persistent phonologic simplifications Fluent, noninformed speech; naming difficulties, phonemic and semantic paraphasia, phonologic deficit, syntactic errors; repetition and oral comprehension impairment; fluent aphasia Hypoinformativity, paraphasia, phonologic deficit, repetition deficit, complex comprehension impairment; poor vocabulary Hypoinformativity, poor vocabulary, impaired elaborate language (inconsistent definitions); written language is acquired; normal academic achievement Mutism Reduced spontaneous speech, anomia, paraphasia; nonfluent aphasia Fluent and informative speech; mild anomia in naming and fluency tests; normal narrative capacity Normal oral speech but reading and spelling difficulties Word finding difficulties in spontaneous speech, paraphasia and perseverations in naming and fluency tests; reading and spelling deficits; low verbal retention; VIQ = 109, PIQ = 85, GIQ = 97; 4th yr of elementary school (stayed down in the 1st yr)
7	2, 0	+	+	+	+	—	—	—	Mutism Anomia, reading difficulties (recognizes vowels only) Mild dysarthria; repetition disorders (for syllables, digits, and nonwords) with auditory comprehension impairment; reduced digit retention; sensorial aphasia Normal except auditory comprehension for long or complex stimuli Normal except nonsemantic items retention (digit span: 4/20); retention of auditory story normal Dysarthria, deficient elaborate language (sentences generation); mild, nonfluent aphasia Normal except elaborate language: poor vocabulary in definition, dysyntaxia, and hypoinformativity in narration; correct naming and fluency tests Mutism for 12 d Slurred and very reduced speech; anomia, phonemic and semantic paraphasia; VIQ = 75, PIQ = 98 Persistent anomia and phonemic paraphasia; poor narrative capacity; written language is acquired Specialized heart disease school Mutism Loss of previous language abilities; says a few isolated words; nonfluent aphasia Severely reduced and agrammatic language, phonologic disorders; normal repetition Normal; some persistent phonologic simplifications Fluent, noninformed speech; naming difficulties, phonemic and semantic paraphasia, phonologic deficit, syntactic errors; repetition and oral comprehension impairment; fluent aphasia Hypoinformativity, paraphasia, phonologic deficit, repetition deficit, complex comprehension impairment; poor vocabulary Hypoinformativity, poor vocabulary, impaired elaborate language (inconsistent definitions); written language is acquired; normal academic achievement Mutism Reduced spontaneous speech, anomia, paraphasia; nonfluent aphasia Fluent and informative speech; mild anomia in naming and fluency tests; normal narrative capacity Normal oral speech but reading and spelling difficulties Word finding difficulties in spontaneous speech, paraphasia and perseverations in naming and fluency tests; reading and spelling deficits; low verbal retention; VIQ = 109, PIQ = 85, GIQ = 97; 4th yr of elementary school (stayed down in the 1st yr)
8	5, 9	—	—	—	—	—	+	+	Mutism Anomia, reading difficulties (recognizes vowels only) Mild dysarthria; repetition disorders (for syllables, digits, and nonwords) with auditory comprehension impairment; reduced digit retention; sensorial aphasia Normal except auditory comprehension for long or complex stimuli Normal except nonsemantic items retention (digit span: 4/20); retention of auditory story normal Dysarthria, deficient elaborate language (sentences generation); mild, nonfluent aphasia Normal except elaborate language: poor vocabulary in definition, dysyntaxia, and hypoinformativity in narration; correct naming and fluency tests Mutism for 12 d Slurred and very reduced speech; anomia, phonemic and semantic paraphasia; VIQ = 75, PIQ = 98 Persistent anomia and phonemic paraphasia; poor narrative capacity; written language is acquired Specialized heart disease school Mutism Loss of previous language abilities; says a few isolated words; nonfluent aphasia Severely reduced and agrammatic language, phonologic disorders; normal repetition Normal; some persistent phonologic simplifications Fluent, noninformed speech; naming difficulties, phonemic and semantic paraphasia, phonologic deficit, syntactic errors; repetition and oral comprehension impairment; fluent aphasia Hypoinformativity, paraphasia, phonologic deficit, repetition deficit, complex comprehension impairment; poor vocabulary Hypoinformativity, poor vocabulary, impaired elaborate language (inconsistent definitions); written language is acquired; normal academic achievement Mutism Reduced spontaneous speech, anomia, paraphasia; nonfluent aphasia Fluent and informative speech; mild anomia in naming and fluency tests; normal narrative capacity Normal oral speech but reading and spelling difficulties Word finding difficulties in spontaneous speech, paraphasia and perseverations in naming and fluency tests; reading and spelling deficits; low verbal retention; VIQ = 109, PIQ = 85, GIQ = 97; 4th yr of elementary school (stayed down in the 1st yr)
9	4, 8	+	+	+	—	+	+	+	Mutism Anomia, reading difficulties (recognizes vowels only) Mild dysarthria; repetition disorders (for syllables, digits, and nonwords) with auditory comprehension impairment; reduced digit retention; sensorial aphasia Normal except auditory comprehension for long or complex stimuli Normal except nonsemantic items retention (digit span: 4/20); retention of auditory story normal Dysarthria, deficient elaborate language (sentences generation); mild, nonfluent aphasia Normal except elaborate language: poor vocabulary in definition, dysyntaxia, and hypoinformativity in narration; correct naming and fluency tests Mutism for 12 d Slurred and very reduced speech; anomia, phonemic and semantic paraphasia; VIQ = 75, PIQ = 98 Persistent anomia and phonemic paraphasia; poor narrative capacity; written language is acquired Specialized heart disease school Mutism Loss of previous language abilities; says a few isolated words; nonfluent aphasia Severely reduced and agrammatic language, phonologic disorders; normal repetition Normal; some persistent phonologic simplifications Fluent, noninformed speech; naming difficulties, phonemic and semantic paraphasia, phonologic deficit, syntactic errors; repetition and oral comprehension impairment; fluent aphasia Hypoinformativity, paraphasia, phonologic deficit, repetition deficit, complex comprehension impairment; poor vocabulary Hypoinformativity, poor vocabulary, impaired elaborate language (inconsistent definitions); written language is acquired; normal academic achievement Mutism Reduced spontaneous speech, anomia, paraphasia; nonfluent aphasia Fluent and informative speech; mild anomia in naming and fluency tests; normal narrative capacity Normal oral speech but reading and spelling difficulties Word finding difficulties in spontaneous speech, paraphasia and perseverations in naming and fluency tests; reading and spelling deficits; low verbal retention; VIQ = 109, PIQ = 85, GIQ = 97; 4th yr of elementary school (stayed down in the 1st yr)

CN = caudate nucleus; D = day; GIQ = Wechsler Global IQ; ICA = internal capsule, anterior limb; ICP = internal capsule, posterior limb; M = month; Pal = pallidum; Put = putamen; SOC = semiovale center; Th = thalamus; VIQ = Wechsler Verbal IQ; Y = year.

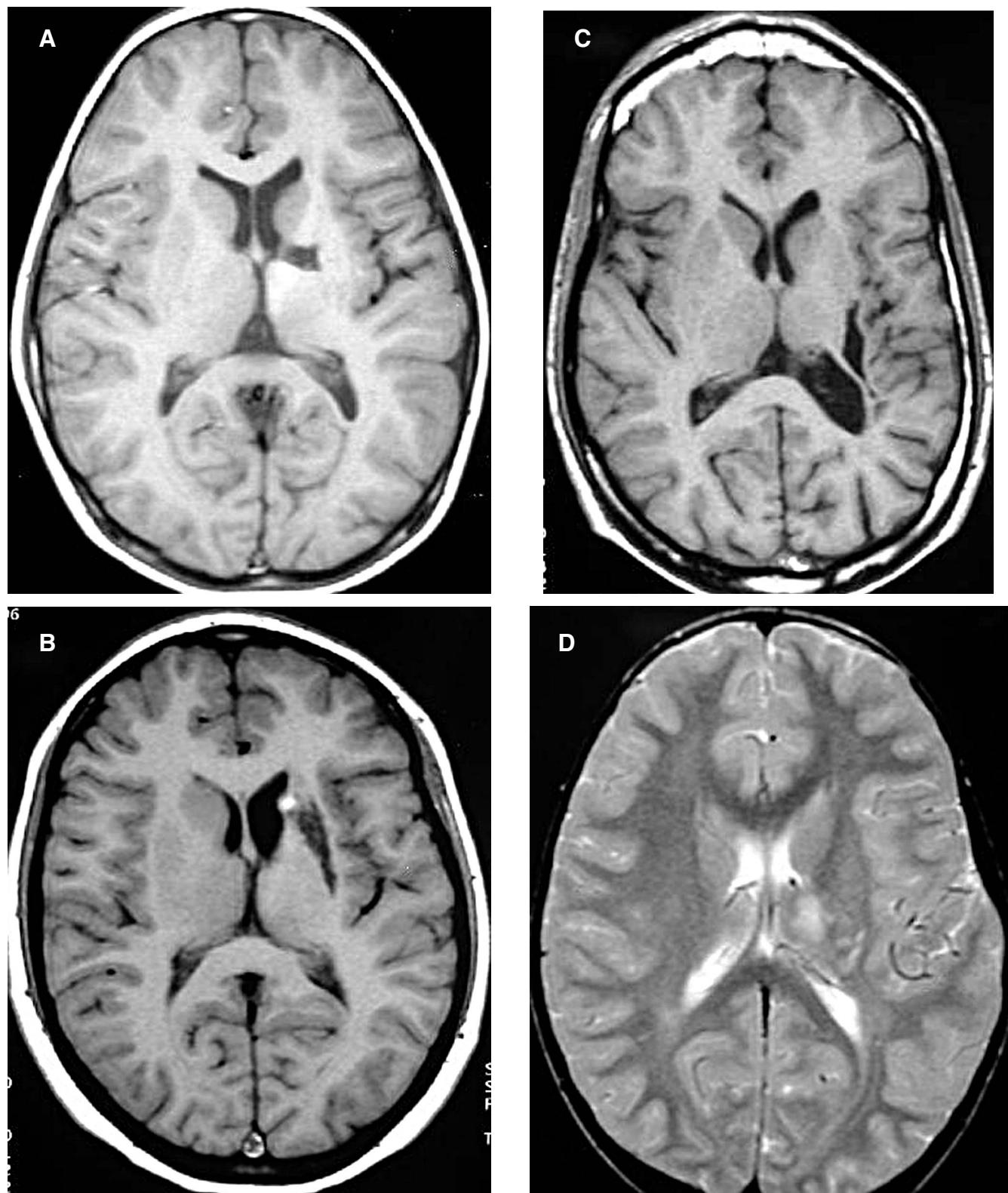


Figure 1. A, Patient 1, magnetic resonance image (MRI) of the brain at 2 years after the infarct: hypointense signal (axial T_1 -weighted spin echo) of the posterior limb of the left internal capsule and pallidum; B, patient 3, MRI at month 3 after the infarct: abnormal signals (axial T_1 -weighted spin echo) of the anterior limb of the left internal capsule, putamen, pallidum, and caudate nuclei; C, patient 4, MRI at year 4 after the infarct: hypointense signal (axial T_1 -weighted spin echo) involving the posterior part of the left internal capsule, putamen, and semiovale center; D, patient 8, MRI at day 9 after the stroke: hyperintense signal (axial T_2 -weighted spin echo) involving the left thalamus, the posterior area of the left striatum, and the posterior limb of the left internal capsule.

Repetition tests showed difficulties in verbal repetition in four children that lasted over time. In patients 1, 4, and 9, difficulties were observed in repetition of low semantic content stimuli (digits, syllables), whereas repetition of sentences and texts was normal; this profile suggests working memory difficulties. In patient 3, memory deficits were detected for sentences and texts, which is in favor of deficit in auditory short-term memory.

Among the eight children reaching school age at the time of the last follow-up, five presented with school difficulties (1-year delay, patients 1, 2, and 9; apprenticeship orientation, patient 3; special needs education, patient 6).

Global psychometric evaluations undertaken in patients 1, 6, and 9 revealed Wechsler Performance and Verbal IQs in the normal range (see Table 1).

Prognostic Factors

In relation to age at the time of stroke, five of the six patients in the preschool group developed language sequelae, whereas in the group of the three older patients, only one child endured sequelae. Concerning the topography of the infarct, among the three patients who had lesions affecting the caudate nucleus, pallidum, putamen, and anterior capsule, two children had a good prognosis. In contrast, among those with posterior lesions (whether isolated or associated), only one of six had a favorable outcome.

DISCUSSION

This study furthers our knowledge of subcortical aphasia and underlines the occurrence of this entity as a complication in deep brain infarcts in childhood. Aphasia was associated with left-hemisphere lesions. Most patients presented with nonfluent aphasia (Broca type); there was also a case of fluent aphasia and a case of sensorial aphasia. Initial language impairments were found to affect mainly expression and included mutism, hypospontaneity, nonfluent speech, and word finding difficulties. Positive signs of aphasia, such as phonemic and semantic paraphasia and neologisms, were frequently observed, as in adult aphasia. Comprehension difficulties were less common. Two thirds of our patients presented with sequelae with a profile of naming difficulties, reduced fluency, a reduction in the lexical stock, repetition difficulties, and difficulties in written language. It is worth noticing that spontaneous and conversational speech seemed quite normal and that standardized tests were needed to highlight signs of aphasia.

Our observations suggest that prognosis is more favorable in children of school age compared with younger patients, who seem to be most vulnerable. In the younger children, the lesions resulted in "delayed" (later appearing) difficulties in written language acquisition; late repercussions of aphasia in early childhood are observed on written language, although oral language is correct. Even when oral language seemed to recover, an alteration of learning potential in abilities such as reading and writing could exist. The cognitive prerequisite for acquisition of written language might be impaired, and the subcortical pathways might play a (direct or indirect) role during the "early" stages of written language learning. Although younger age at the time of a neurologic event is usually considered a factor of better prognosis,¹⁶ stroke was already reported as a significant predictor of adverse outcome in childhood¹⁷ and impairment in learning new language mediated skills was reported.⁶

Intellectual functions were relatively preserved at both the initial phase and later; when patients had an intellectual assessment, IQ scores were normal or subnormal. Most patients (in our series) returned to school, and even when there was a 1-year delay, they continued with standard courses.

Although cognitive impairments as a consequence of stroke have already been reported in childhood,¹⁷⁻²⁰ few studies have focused on detailed neurolinguistic descriptions of subcortical aphasia after ischemic stroke. The few patients who underwent formal and detailed language assessment ($n = 4$, to our knowledge) were between 5 and 7 years old at the time of the stroke.²¹⁻²⁴ Their speech anomalies were comparable to those observed in our patients and included mutism, followed by low fluency, naming difficulties, and paraphasia. Anomalies of verbal comprehension were transitorily seen in two patients. Verbal repetition was impaired in two of them. Most often phonologic and syntactic difficulties were absent. The prognosis was favorable in three children. However, the follow-up of less than 6 months would not allow comprehensive evaluation of long-term prognosis and would therefore not rule out the risk of delayed sequelae. The fourth patient had persisting sequelae at the last evaluation, 6 years after the infarct.²⁴ Initially, this patient showed a low fluency and moderate oral comprehension disorders. Six years later, these difficulties persisted and were associated with a spelling disorder, dyslexia, and poor school grade.

Acquired aphasia in children was also described after surgical resection of left thalamic tumors²⁵; localizations were not strictly identical, and the profile of aphasia was close but more heterogeneous: three patients had Broca aphasia, one had mixed transcortical aphasia, and one had conduction aphasia.

In adulthood, for many years, studies reported speech disorders following the occurrence of basal ganglia lesions.²⁶⁻²⁹ Some patients were dysarthric. Regarding aphasic symptoms, there was, as in our young patients, a predominance of expression disorders, with slow fluency, naming difficulties, and paraphasia. Difficulties in comprehension were rarely observed. Most of these speech disorders did not seem to fully recover. Written language impairment results from different mechanisms in adults than those in children: alexia and agraphia concern acquired abilities, whereas in children, written language is still being acquired.

Functional imaging techniques are of particular interest in studying the involvement of the basal ganglia in language processing, especially after damage. Dynamic studies after subcortical or thalamic insult in adults, showing cortical hypometabolism, suggest that the basal ganglia could be an integrator of language functions via verbal memory or semantic mechanism and could be an activator of cortical language functions.²⁵

To conclude, our observations, supported by those of others, stress the importance of the basal gray nuclei and the adjacent white-matter pathways in the development and use of oral and written language. In children, these subcortical neuronal centers and fiber pathways appear to be implicated particularly in the expression of speech, especially in vocabulary access and storage.

From a practical point of view, our results clarify the neurolinguistic approach to be adopted when faced with subcortical lesions involving an area potentially dedicated to linguistic functions in children. An early and formal language assessment is required to investigate both expressive and receptive aspects. This initial assessment should be followed by sequential examinations

to guide rehabilitation and to detect the occurrence of secondary disorders, especially in the field of written language acquisition and memory.

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