

Acquired Capsular/Striatal Aphasia in Childhood

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● We studied a case of language loss caused by an acquired vascular lesion in the putamen, anterior limb of the internal capsule, and lateral aspect of the head of the caudate nucleus in a 7-year-old right-handed girl. Acute right-sided hemiplegia, mutism, oral apraxia, and disturbance in language comprehension but no dysarthria were present. During recovery, a nonfluent aphasia with anomia was evident. After six months, only mild hemiparesis and minor spelling difficulties persisted. We compared this patient with an 11-year-old right-handed girl with right-sided hemiparesis and dysarthria but no language loss following a lesion in the globus pallidus, a portion of the posterior limb of the internal capsule, and the body of the caudate. The presence of a language disturbance in the first but not the second patient was attributed to the difference in lesion location. The symptoms and lesions were similar to those in recent reports of adult patients. To our knowledge, this is the first report of these findings in a child with a left-hemisphere lesion.

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Language and articulatory disturbances caused by lesions in the basal ganglia and internal capsule have been reported in adult patients.¹⁻⁸ Primary among the features generally described have been relatively persistent hemiparesis, dysarthria, and language disturbances unlike those in cortical aphasic syndromes. Damasio et al¹ described six cases of atypical aphasia caused by nonhemorrhagic lesions in the dominant basal ganglia and internal capsule, comparing the language findings with those for four patients with similar lesions in the nondominant basal ganglia who did not have an aphasia. Naeser et al² detailed language and articulatory findings for nine patients, all with dominant-hemisphere capsular/putaminal lesions, suggesting that three subcortical aphasic syndromes according to lesion site are identifiable.

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In a recent study Brunner et al³ described eight relatively persistent cases of aphasia from left-hemisphere basal ganglia lesions. Language abnormalities were not detected in a second group of patients who sustained left-hemisphere white-matter lesions in the vicinity of the basal ganglia but not involving these nuclei. They did not detect language abnormalities in patients with basal ganglia lesions in the right hemisphere.

Our report describes an unusual case of acquired capsular/striatal aphasia in a 7-year-old girl, comparing these findings with those in an 11-year-old girl with an acquired lesion in the globus pallidus, posterior limb of the internal capsule, and body of the caudate who did not have language loss.

REPORT OF CASES

CASE 1.—A 7-year-old right-handed girl had an acute left-sided temporal headache followed by slurred speech and right-sided weakness. Within ten minutes she was unable to speak and was taken to a suburban hospital in a semiconscious state with pronounced hemiplegia on the right. On day 2, because of moderate systolic hypertension in the right arm and suspected coarctation of the aorta, she was transferred to Rainbow Babies and Childrens Hospital, Cleveland. At that time she was mute, although alert, with right-sided hemiparesis affecting the arm and face more than the leg.

When first seen for a language consultation on day 4, the child was completely mute and unable to initiate nonspeech oral movements on command, such as blowing or yawning, although she could imitate these nonspeech oral movements successfully. Language comprehension was moderately impaired, falling 1½ years below age expectations on a test of vocabulary comprehension, the Peabody Picture Vocabulary Test,⁹ and at a 4½-year-old level in her understanding of syntactic contrasts, as estimated by the Northwestern Syntax Screening Test: Receptive Portion.¹⁰ The patient was able to identify simple printed words, including her name, *mom*, *go*, *yes*, and *no*, demonstrating considerable reading recognition for a child who had just entered second grade. By the sixth day, voluntary initiation of phonation and voluntary production of nonspeech oral movements were achieved on command. By discharge on day 8, the hemiplegia had resolved to moderate weakness in the right arm and mild weakness in the right leg. The patient could protrude, retract, and lateralize her tongue on command, as well as place her upper teeth on

her lower lip. She also was able to imitate vowel sounds and spontaneously began to initiate short agrammatic phrases such as *hi*, *Barb* and *go home*. A full range of tongue movements was present, although the mild right-sided facial paresis caused lip movements to be asymmetric. The little speech used was not noticeably dysarthric. She remained severely anomic, completely unable to name on confrontation.

One month ten days after the vascular lesion, the patient had mild weakness in the right arm and leg. Language comprehension had returned to normal, with vocabulary comprehension, as assessed by the Peabody Picture Vocabulary Test,⁹ at a 9-year 2-month age level. Receptive syntax was assessed by the Northwestern Syntax Screening Test¹⁰ at the 50th percentile for children her age. Expressive language, although improved, was principally characterized by short, occasionally agrammatic sentences with continued semantic paraphasias (eg, *stir up my bed* for *crank up my bed*) and word-finding difficulties (eg, *thing that goes around for propeller*). On the Sentence Repetition Test¹¹ she was unable to repeat correctly approximately one third of the sentences, making syntactic errors in several contexts.

Two months, six days after the incident, results of the Wechsler Intelligence Scale for Children—Revised (WISC-R)¹² were as follows:

	Scaled Score
Verbal tests	
Information	12
Similarities	11
Arithmetic	12
Vocabulary	10
Comprehension	10
Digit span	13
Performance tests	
Picture completion	13
Picture arrangement	12
Block design	16
Object assembly	15
Coding	6

The verbal IQ was 106; performance, 117; and full-scale, 112. Because of residual right-sided hemiparesis, the child continued to perform writing tasks with her left hand, thus impairing speed, which was considered the chief factor for the depressed coding score on the WISC-R. Spontaneous expressive language at this time was predominantly simple sentence structures (62% of spontaneous speech sample) and elementary conjunctions (*and*, *and then*) with no use of sentence subordination. However, the patient had no difficulty identifying and correcting ungrammatical sentences when given the Syntactic Errors Test.¹¹

Six and one-half months after onset, the right-sided hemiparesis resolved to a mild residual clumsiness of the right hand. All

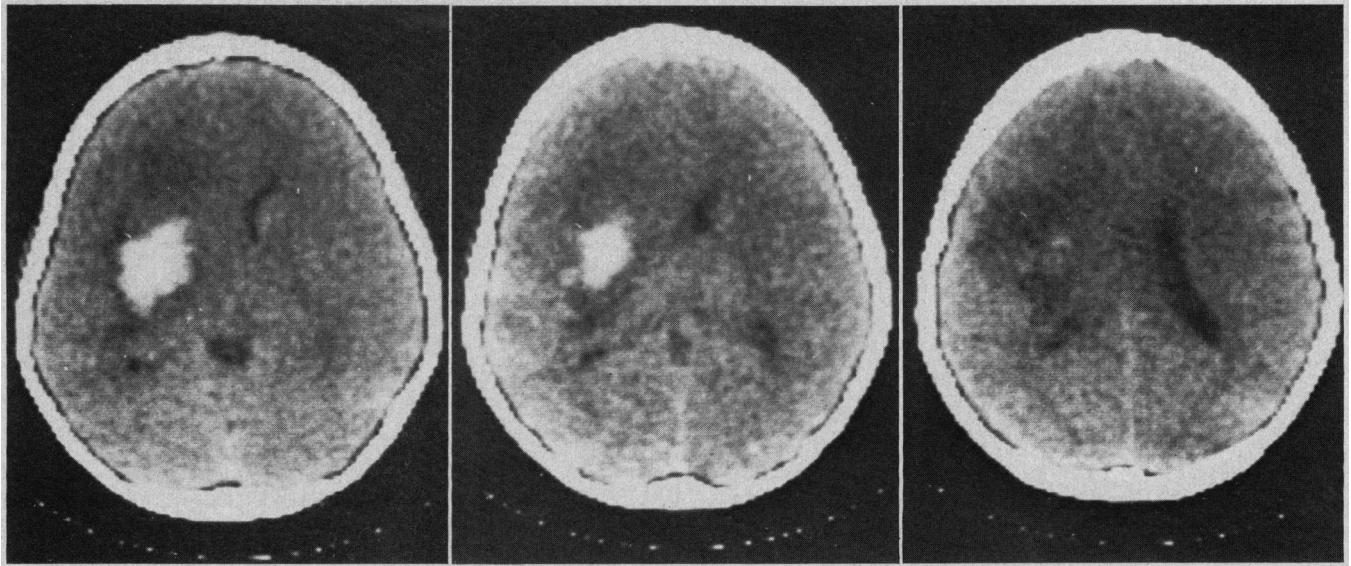


Fig 1.—Computed tomographic scan without contrast (case 1) performed two days after lesion's occurrence shows area of increased and decreased density in depth of left hemisphere.

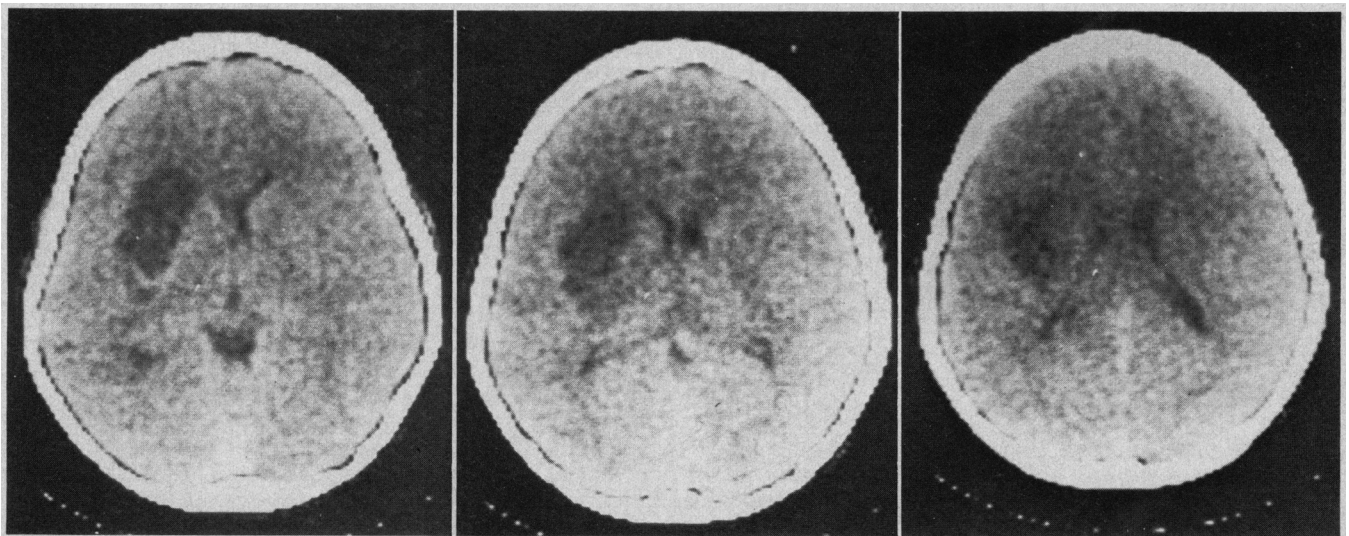


Fig 2.—Computed tomographic scan (case 1) at one month shows area of decreased density in depth of left hemisphere.

verbal language abilities recovered completely, as shown by formal testing and spontaneous language samples. The patient's reading comprehension, word attack, mathematical calculations, and applied problems were at her grade level, despite one half of a year's absence from school because of the illness. Written language skills, including spelling, continued to be depressed by one grade level.

The computed tomographic (CT) scan done on admission showed an area of increased density in the depth of the left hemisphere, involving the entire putamen. A contiguous, low-density area extended anteriorly across the anterior limb of the internal capsule and involved the white matter just anterior and lateral to the

frontal horn of the lateral ventricle. The lateral aspect of the head of the caudate may also have been involved (Fig 1). A repeated CT scan at one month revealed an area of decreased density closely corresponding to previous areas of increased and decreased densities (Fig 2). Arteriograms performed initially on admission and at one month did not show an aneurysm or arteriovenous malformation. Cardiac catheterization performed one month after the lesion's occurrence revealed a bicuspid aortic valve and coarctation of the aorta.

CASE 2.—An 11-year 8-month-old right-handed girl had a sudden temporal headache on the left followed by right-sided hemiparesis. Within minutes, she became

lethargic and mute. On admission to Rainbow Babies and Childrens Hospital several hours after onset, she was alert and oriented, although dysarthric, with right-sided hemiparesis affecting the arm more than the face and leg. The patient also had hypoesthesia and decreased position sense over the right arm. At 24 hours, repetition and reading abilities were intact, and she could follow simple three-step commands. After one week, sensation was normal, although moderate right-arm and mild right-leg weakness remained. At that time, the only apparent speech disorder was a very mild dysarthria characterized by occasional consonant cluster reductions (eg, *scatch* for *scratch*) and consonant omissions in multisyllabic words (eg, *info-*

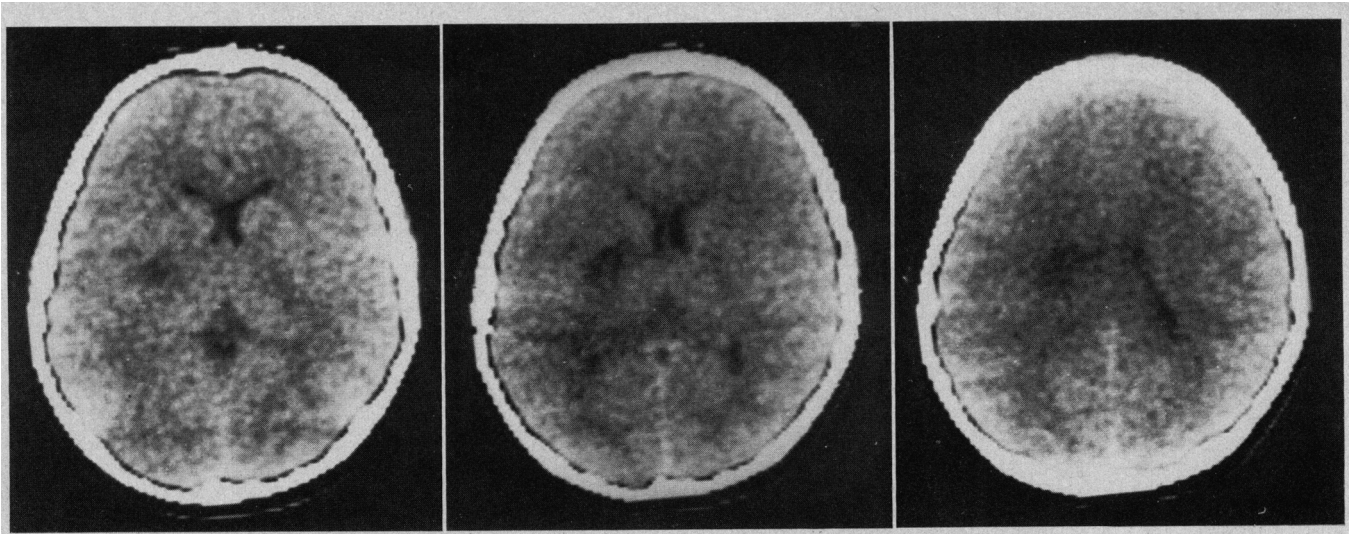


Fig 3.—Computed tomographic scan (case 2) performed on day 6 shows area of decreased density in depth of left hemisphere.

ration for information). Normal language comprehension and expression were documented. One year after onset of symptoms, no dysarthria was noted, although a residual mild weakness in the right hand remained.

A CT scan showed an area of decreased density in the depth of the left hemisphere (Fig 3), involving the posterior aspect of the globus pallidus, posteromedial aspect of the putamen, a portion of the posterior limb of the internal capsule, and the body of the caudate. An arteriogram revealed narrowing of the supraclinoid portion of the left internal carotid artery and a beaded appearance of the horizontal portion of the left middle cerebral artery, suggesting intracranial fibromuscular dysplasia.

COMMENT

Acutely, patients 1 and 2 had strikingly different clinical appearances, the first involving language comprehension and expression but not articulation and the second involving articulation but not language.

Case 1 represents the only known reported case of language loss following a capsular/striatal lesion in the left hemisphere in a child. Acute findings were similar to those in cases 1, 4, 5, and 6 reported by Damasio et al¹; his patients had acute symptoms of hemiplegia involving the arm more than the leg or face, nonfluent aphasias, phonemic and semantic paraphasias, and difficulties with comprehension, repetition, and naming. Unlike these four patients, our patient 1 was not dysarthric. The lesion site in those four patients involved the body and somewhat more of the head of the caudate but did not extend as far posteriorly or laterally in the putamen. Our patient 1 was also similar to

three patients of Naeser et al² with capsular/putaminal lesions with anterior and superior extension, again with the exception that our patient was not dysarthric. The three patients of Naeser et al had right-sided hemiparesis affecting the arm more than the leg, dysarthria, nonfluent aphasia, somewhat impaired verbal comprehension, and impaired repetition and naming ability. Computed tomographic scans in their first three patients showed involvement of the putamen, anterior limb of the internal capsule, and probably a variable involvement of the head and body of the caudate.

In comparison with Alexander and LoVerme's⁴ six patients with left-hemisphere hemorrhagic lesions with putaminal focus, our patient 1 had initial symptoms similar to those of their patients 10 and 14, who had mild or no dysarthria; mild to moderate impairment of comprehension and repetition; moderate to severe impairment of naming, reading, writing ability; and paraphasias. However, both of those patients were fluent at the time of testing. Their lesion sites were similar to those of our first patient, although the CT reconstructions did not include subcortical landmarks and did not permit more precise comparison of the structures involved. Only one of the eight cases of Brunner et al³ exhibited (mild) dysarthria; these patients all had a lenticular focus and are most comparable with our patient 1, with language comprehension somewhat better preserved than expression. One of their patients, described in some detail, sustained an infarct in the body of the caudate,

internal capsule, and globus pallidus. Eight months later this patient still had anomia, phonemic and semantic paraphasias, and other aphasic sequelae.

Our patient 2 is similar to patient 7 of Damasio et al¹ in that both had initial right-sided hemiparesis affecting the arm more than the face and leg and dysarthria without aphasia. Both patients had lesions involving the midportion of the body of the caudate, adjacent white matter of the corona radiata, and the medial aspect of the putamen. Patient 2 is somewhat similar to Fisher's⁵ patient 7, who had initial right-sided hemiparesis and a severe dysarthria but also some word-finding difficulties. The lesion involved the superior portion of the genu of the internal capsule and body of the caudate and extended laterally through the lateral putamen. However, the lesion also extended somewhat anteriorly, involving the anterior limb of the internal capsule.

The only other reported case of a child with a basal ganglia lesion for whom CT localization and language testing were done was a 6-year-old left-handed girl with a right-hemisphere lesion.¹³ Her clinical appearance and CT scan lie somewhere between our two cases, although the lesion was in the right hemisphere. She was presumed to be right hemisphere dominant for language, although her handedness was questionable. Like our patient 1, she was nonfluent and had difficulty with repetition and some difficulty with naming. Like patient 2, she had dysarthria and normal comprehension. Her lesion site did not extend as far anteriorly as

patient 1's and did not involve either the anterior limb of the internal capsule or head of the caudate. However, her lesion extended somewhat farther laterally than patient 2's and probably involved a greater portion of the putamen.

In contrast to comparable adult cases, our two pediatric patients recovered more rapidly and completely. By one month after onset, our first patient had regained normal comprehension and was speaking in grammatical but elementary sentences, with only a mild residual anomia. By six months, only clumsiness in the right hand and a mild spelling difficulty persisted. In contrast, patients 1, 4, 5, and 6 of Damasio et al,¹ patients 1, 2, and 3 of Naeser et al,² patients 10 and 14 of Alexander and LoVerme,⁴ and eight patients of Brunner et al³ showed only gradual improvement over several months and continued to have residual defects up to several years later. By two weeks, our patient 2's dysarthria had resolved whereas patient 7 of Damasio et al¹ continued to be dysarthric three months after the lesion's occurrence. The 6-year-old left-handed patient with a right-hemisphere lesion described by Ferro et al¹³ also rapidly recovered language, although because of her writing ambidexterity, partial bilateral representation of language could not be ruled out.

The differences in clinical appearance between our two patients may well be the result of differences in lesion location. The region in the left hemisphere including the head of the caudate, anterior limb of the internal capsule, and anterior and superior aspect of the putamen has repeatedly been implicated in a syndrome that

includes aphasia, right-sided hemiparesis, and, often, dysarthria.¹⁻⁸ These structures were involved in patient 1, who had acute language abnormalities, but not in patient 2, whose language was never disturbed.

The role of each of these structures in language, however, has yet to be specified by descriptions of patients with lesions confined to single structures and by neuropathologic verification of lesion sites. An isolated hemorrhage of the head of the left caudate has been reported.⁶ The patient had no comprehension difficulties, no dysarthria, and no phonemic paraphasia, yet speech and graphic expression were "incoherent" and characterized by "perseveration of ideas, semantic inconsistencies and inability to keep a stable objective." Although this might indicate the anterior limb of the internal capsule or anterior/superior putamen as responsible for these comprehension deficits, a patient with an isolated lesion in either of these regions has yet to be described.

A neuropathologic study has been reported⁷ for a patient who sustained an infarct in the body and upper part of the head of the caudate, the entire putamen, adjacent superior part of the internal capsule, and anterior nucleus of the thalamus. This right-handed patient had fluent aphasia characterized by perseverations and semantic and ideation incoherences. Finally, recent animal studies reviewed extensively by Damasio et al¹ have shown prominent corticostriatal inputs from the auditory and premotor cortices to the head of the caudate by way of the anterior limb of the internal capsule.^{14,15} In addition, the white matter anterior to the frontal horn of the lateral ventricle has been

found to be traversed by reciprocal projections between the face region of the supplementary motor area and premotor cortices.¹⁶ These studies were done in rhesus monkeys, but similar tracts are thought to occur in man.

Since the major neural output of two of the three structures in this area (caudate and putamen) is to the globus pallidus and then to the lateral ventral nucleus of the thalamus, further studies should compare the language deficits found in capsular/striatal lesions with those found in pallidal/thalamic lesions. Dysphasia following stereotactically placed lesions in the globus pallidus has been reported,¹⁷⁻¹⁹ as has a syndrome involving anomia, dysarthria, dysphonia, and intact comprehension caused by stereotactic lesions of the lateral ventral thalamus.²⁰

The initial mutism and comprehension deficit with semantic paraphasias, anomia, and oral apraxia in case 1 appear further to implicate capsular/striatal structures in the brain's language system. Patient 1's rapid and near-complete recovery suggests that early capsular/striatal lesions may be well compensated for by other brain structures. The absence of aphasia in patient 2, in comparison with adults with lesions of the globus pallidus, may be secondary to limited involvement of these structures by our patient's lesions or may reflect their developmental plasticity. Thus, in addition to describing the role of capsular/striatal structures in language, further studies should consider their developmental plasticity.

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