Regional Brain Blood Flow in Congenital Dysphasia: Studies with Technetium-99m HM-PAO SPECT

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Congenital dysphasia is a developmental speech disorder not explained by deafness, phonation disorder, mental retardation, neurologic lesion, or psychiatric disease. The existence of brain lesions has often been postulated but conventional investigations fail to demonstrate any cerebral abnormality. By means of [^{99m}Tc]hexamethyl-propyleneamine oxime (HM-PAO) brain single photon emission computed tomography (SPECT) we have studied 14 children suffering from congenital dysphasia. The brain computed tomographic scan was normal in all cases. In two patients with expression impairment the SPECT study demonstrated a hypoperfusion in the inferior frontal convolution of the left hemisphere, involving the Broca's area. In nine of 12 patients with global dysphasia (deficits in both comprehension and expression), SPECT study showed two hypoperfused areas: an abnormality in the left temporoparietal region and a hypoactivity in the upper and middle areas of the right frontal lobe. These results suggest that congenital dysphasia could be due, like acquired aphasia, to specific impairment of the language cerebral areas and that brain SPECT studies with [^{99m}Tc]HM-PAO could be useful for a better comprehension of the physiopathology of these disorders.

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Speech disorders are frequently encountered in children (*1,2*). In some cases, these disorders are the result of defects of peripheral speech organs, or of the auditory or central nervous systems. In others, they constitute one of the manifestations of a more complex disorder such as mental retardation or psychiatric disease. There are cases, however, where no causal factor can be found and one speaks about "specific developmental speech disorders" or about "congenital or developmental dysphasia" (*2*). The etiology of these disorders has been long debated. It was previously considered that congenital dysphasia had a psychologic origin. The current view is that congenital dysphasia is in most cases the result of organic cerebral lesions. The nature of the cerebral abnormalities, however, is still unknown. The computed tomographic (CT) scan of the head is usually normal and there is no neuropathologic evidence that the cerebral areas concerned with speech are involved (*1,3*).

Recently, tracers that cross the normal blood-brain barrier and are taken up proportionally to cerebral blood flow have been synthesized. Some of these molecules, like iodine-123 (^{123}I) iodoamphetamine or technetium-99m (^{99m}Tc) hexamethyl-propyleneamine oxime (HM-PAO), can be used with the single photon emission computed tomography (SPECT) facilities available in most nuclear medicine departments. The value of this method has been established in a variety of neurologic disorders in adults and children even when the other procedures such as electroencephalogram and CT scan of the head fail to demonstrate any abnormality (*4–7*).

We report our initial experience of [^{99m}Tc]HM-PAO brain SPECT in children with congenital dysphasia.

PATIENTS AND METHODS

According to a protocol accepted by our local committee for medical ethics, we have investigated, after informed con-

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TABLE 1
Neuropsychologic Data and SPECT Findings of 14 Children with Congenital Dysphasia

Patient no.	Neuropsychologic problems	Technetium-99m HM-PAO SPECT findings
1	Verbal dyspraxia	Inferior left frontal hypoperfusion, involving the Broca's area.
2	Verbal dyspraxia	Inferior left frontal hypoperfusion, involving the Broca's area.
3	Global dysphasia Ideomotor apraxia Reduced prosody	Hypoperfusion in the posterior and superior part of the left temporal lobe. Right upper frontal hypoactivity.
4	Global dysphasia Ideomotor apraxia Reduced prosody	No significant asymmetry.
5	Global dysphasia Attention disturbances Ideomotor apraxia Reduced prosody	Hypoperfusion in the posterior and inferior part of the left parietal lobe. Right upper frontal hypoactivity.
6	Global dysphasia Attention disturbances Ideomotor apraxia Reduced prosody	Hypoperfusion in the posterior and superior part of the left temporal lobe. Hypoactivity in the upper and middle areas of the right frontal lobe.
7	Global dysphasia Attention disturbances Ideomotor apraxia Reduced prosody	No significant asymmetry.
8	Global dysphasia Ideomotor apraxia Reduced prosody	Hypoperfusion in the posterior and superior part of the left parietal lobe. Right upper and middle frontal hypoactivity.
9	Global dysphasia Attention disturbances Ideomotor apraxia Reduced prosody	No significant asymmetry.
10	Global dysphasia Attention disturbances Reduced prosody	Hypoperfusion in the posterior and superior part of the left temporal lobe. Hypoactivity in the upper and middle areas of the right frontal lobe.
11	Global dysphasia Attention disturbances Reduced prosody Ideomotor apraxia	Hypoperfusion in the posterior and inferior part of the left parietal lobe. Right upper frontal hypoactivity.
12	Global dysphasia Attention disturbances Ideomotor apraxia Reduced prosody	Hypoperfusion in the posterior and superior part of the left temporal lobe. Right upper frontal hypoactivity.
13	Global dysphasia Ideomotor apraxia Reduced prosody	Hypoperfusion in the posterior left temporoparietal lobe. Hypoactivity in the upper and middle areas of the right frontal lobe.
14	Global dysphasia Attention disturbances Ideomotor apraxia Reduced prosody	Hypoperfusion in the posterior left temporoparietal lobe. Hypoactivity in the upper and middle areas of the right frontal lobe.

sent of the parents, 14 patients (ten males, four females), aged 5 to 16 yr, suffering from congenital dysphasia. A familial history of speech disorders was reported in five patients and a birth asphyxia in two. Autism and deafness could be excluded in all cases. Neurologic examination was normal or revealed only some aspecific "soft" signs, such as moderate impairment in standing balance, coordination or gait. The electroencephalogram was moderately too slow in one child and strictly normal in all the other patients. The CT scan of the head, performed with the Somatom Siemens DRH head scanner, was normal in all children.

Neuropsychological Data

The language and the other neuropsychological functions were evaluated by direct clinical observation and by a battery of standardized tests for French speaking children older than 4 yr (verbal auditory comprehension: execution of verbal consigns, WISC-R, Comprehension and Vocabulary subtests; phonology: repetition of easy and complex words, naming of common objects; syntax, articulation and fluency: analysis of spontaneous speech production; intellectual performances: Ravens Progressive Matrices; attention: Continuous Visual Reaction Time Test; ideomotor praxia: reproduction of Rey Complex Figure, copy part) (*8*).

Two children had an impaired expression (verbal dyspraxia according to the Rapin and Allen criteria). The other 12 children had a global dysphasia (deficiencies in both comprehension and expression-phonologic syntactic disorder, according to the Rapin and Allen criteria) (*9*). In addition to their

language disorder, attention disturbances, ideomotor apraxia, and reduced prosody (intonation of speech) were, respectively, present in eight, 11, and 12 children with global dysphasia. No patient was mentally retarded (Table 1).

Technetium-99m HM-PAO SPECT Study

The child was placed in a quiet environment and an i.v. line was then inserted. A few minutes later 0.5 mCi/kg of [^{99m}Tc]HM-PAO was administered. No premedication was given. SPECT imaging was performed 10 to 60 min after i.v. injection using an Elscint rotating gamma camera and a low-energy, high resolution collimator. Data for 360° were collected using 30-sec frames and 6-degree increments. Transaxial, coronal, and sagittal reconstructions were calculated after a high frequency cutoff using a Hamming-Hann filter by an Apex 415 computer system. Slices were 2 pixels thick (0.5 to 1 cm). Only differences of more than 12% between symmetric regions of the brain were considered significant according to our experience in children suffering from various neurologic disorders.

RESULTS

In the two patients with expression impairment the SPECT study demonstrated a hypoperfusion in the inferior frontal convolution of the left hemisphere, involving the Broca's area.

In nine of 12 patients with global dysphasia (deficiencies in both comprehension and expression), the SPECT study showed two hypoperfused areas: an abnormality in the left temporoparietal region and a hypoactivity in the upper and middle areas of the right frontal lobe. The degree and the extent of the hypoperfusions varied however from one patient to another. In the three other patients no significant abnormality was demonstrated.

Results are detailed in Table 1. Examples of SPECT images obtained in dysphasic children are shown in Figure 1.

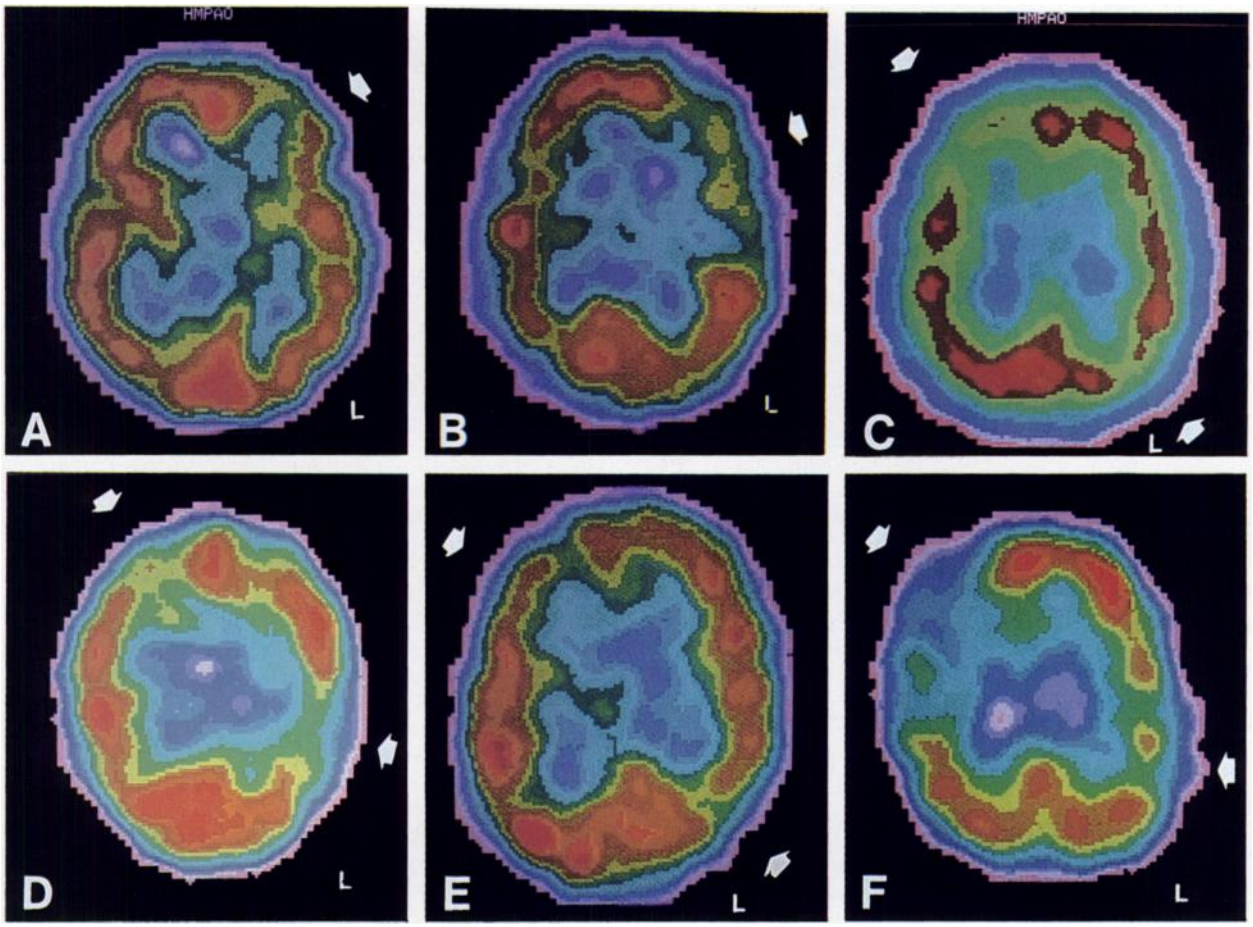

FIGURE 1
Transaxial SPECT images in six children (A to F) suffering from congenital dysphasia. L indicates the left side. The SPECT abnormalities are shown by the arrows. Slice level is ~3 cm above the orbitomeatal line in A and B, ~5 cm in C, E, F, and ~7 cm in D. A (case 1) and B (case 2): Patients with expression impairment: hypoperfusion in the inferior frontal lobe of the left hemisphere, including the Broca's area. C to F: Patients with global dysphasia (cases 6, 8, 13, 14): hypoperfusion in the posterior part of the left temporo-parietal lobe and right frontal hypoactivity.

DISCUSSION

Congenital dysphasia is a developmental language disorder not explained by deafness, phonation disorder, mental retardation, neurologic lesion, or psychiatric disease. This disorder differs from acquired aphasia because of the existence, in many cases, of reduced prosody, ideomotor apraxia, and/or attention disturbances and because of the absence of demonstrable structural abnormality (*1,3*).

Technetium-99m-labeled radiotracers for cerebral blood flow study with SPECT have been synthesized recently. This method is highly attractive because of its convenience compared to positron emission tomographic imaging techniques and its capacity to detect regional abnormalities not visualized by other techniques (*4–7*). To obtain good quality images, the administered activities in children are, on a per kilo basis, higher than in adult patients. The absorbed dose remains, however, in the usual range of that delivered by other radionuclides studies using ^{99m}Tc-labeled molecules (*10*).

In the present work, using [^{99m}Tc]HM-PAO brain SPECT, language areas hypoperfusions have been noticed in 11 out of our 14 children suffering from congenital dysphasia. The hypofixation found in the Broca's area could explain the expression impairment in the two patients with verbal dyspraxia. In patients with global dysphasia, the existence of a hypoperfusion in the left temporoparietal area could explain the difficulties in the comprehension of speech. However, as they also had difficulties in the verbal expression, the absence of lesion in the Broca's area is noteworthy. This finding suggests, in agreement with some neuropsychologic studies, that in congenital dysphasia the abnormal expressive speech is frequently the consequence of impaired comprehension (*11–14*).

The presence of a hypoperfusion of the right frontal lobe in patients with language disorder is rather unexpected. These anomalies are probably not related to the dysphasia itself but to the associated deficits more particularly to the reduced prosody and the attention disturbances. As a matter of fact, such deficiencies are specifically encountered in right-hemispheric damaged children (*15–17*).

In three of our 12 children with global dysphasia the SPECT was normal. These patients could have similar but smaller and undetectable abnormalities. They could also belong to another subgroup of patients, even if the retrospective analysis of their clinical data did not reveal obvious differences with the group of patients whose SPECT study was abnormal.

Till now, only a few scintigraphic studies of congenital dysphasia are available. Lou et al., using SPECT and xenon-133 (^{133}Xe) have found in eight patients with congenital dysphasia symmetrical hypoperfusions in the perisylvian regions. Abnormalities were located anteriorly in expressive dysphasia, posteriorly in comprehension impairment and both anteriorly and posteriorly in global dysphasia (*18*). However, most of the children studied by these authors had a history of neonatal problems which could explain the distribution of the abnormalities in the watershed areas of the major cerebral arteries. Other authors, also using SPECT and ^{133}Xe, have found in 11 children with expressive dysphasia a posterior temporal lesion (unilateral or bilateral) in all patients, a posterior parietal defect in five and a frontal hypoactivity in four (*19*). These results suggest, in agreement with our findings, that an impaired expression can be observed without lesion of the Broca's area and can be the result of a more posterior defect.

In conclusion, our preliminary study shows that some children suffering from congenital dysphasia can have regional cerebral blood flow abnormalities involving the language areas. Since congenital dysphasia artificially regroups all patients with developmental language disorders of unknown origin, these results also suggest that brain SPECT studies with [^{99m}Tc]HM-PAO could be useful for a better comprehension of the physiopathology of these disorders.

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