Case Report

N-ISOPROPYL-P-^{123} I-IODOAMPHETAMINE SPECT AND MAGNETIC RESONANS OF THE BRAIN IN A CASE WITH HYPERGLYSINEMIA

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SUMMARY: Presented here is a case with hyperglysinemia who underwent CT, IMP SPECT and MR imaging. The modalities (CT and MRI) which image the brain structure failed to explain the main neuropsychological defect. However, the IMP SPECT study showed an abnormal function map which correlated well with the clinical findings and it was concluded that: IMP SPECT was more sensitive and informative than the other imagine modalities to reveal even a mild brain damage secondary to the metabolic disorders.

Key Words: Hyperglysinemia, CT, MR Imaging, IMP SPECT.

INTRODUCTION

Metabolic diseases of childhood may cause mental retardation, seizure, motor deficit and ataxia. These may primarily involve the white matter, gray matter or both gray and white matter regions (3). The earlier is the detection, the less damage to the CNS and, therefore, appropriate diagnostic modalities are crucial for early diagnosis which can be helpful to prevent further neurological damage especially in the childhood.

This report is the analysis of the results of Computerized Tomography (CT), Magnetic Resonance (MR) Imaging and N_Isoprophyl-pl^{123}-iodoamphetamine single photon emission tomography (IMP SPECT) of the brain in a case with hyperglysinemia.

CASE REPORT

A ten month old female baby was brought to the Fukui University Hospital, Department of Neurosurgery following several days of agitation, left hemiplegia, fever, ill temper and vomiting. The family reported that the baby remained healthy until approximately 7 months of age when a gradual onset of development delay was noticed. She vomited more than 3 times a day before admission to the hospital.

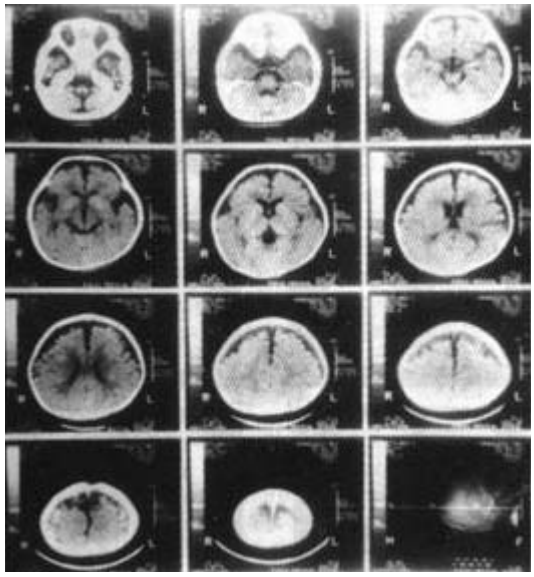

Figure 1: X-ray computerized tomographic scan of a 10 month old baby showing mild cortical atrophy and enlarged subdural space in the both frontal labes.

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There were no remarkable problems during pregnancy or delivery. The physical examination releaved bulged fontanel, neck rigidiy and left hemiplegia. Routine blood biochemistry examination was normal. The result of arterial blood gases revealed metabolic acidosis and respirartory compensation. CT examination was performed immediately, revealing a relatively large frontal subdural space and slight frontal atrophy bilaterally (Figure 1), MR (T_1 and T_2 weighted spin echo pulse sequence) demonstrated no additional abnormality (Figures 2a, b). The findings of digital Substraction Angiography (DSA) was normal (Figures 3a, b). For further investigation, IMP SPECT study was ordered. SPECT was performed with a rotating gamma camera (GE 400AC/STARCAM)

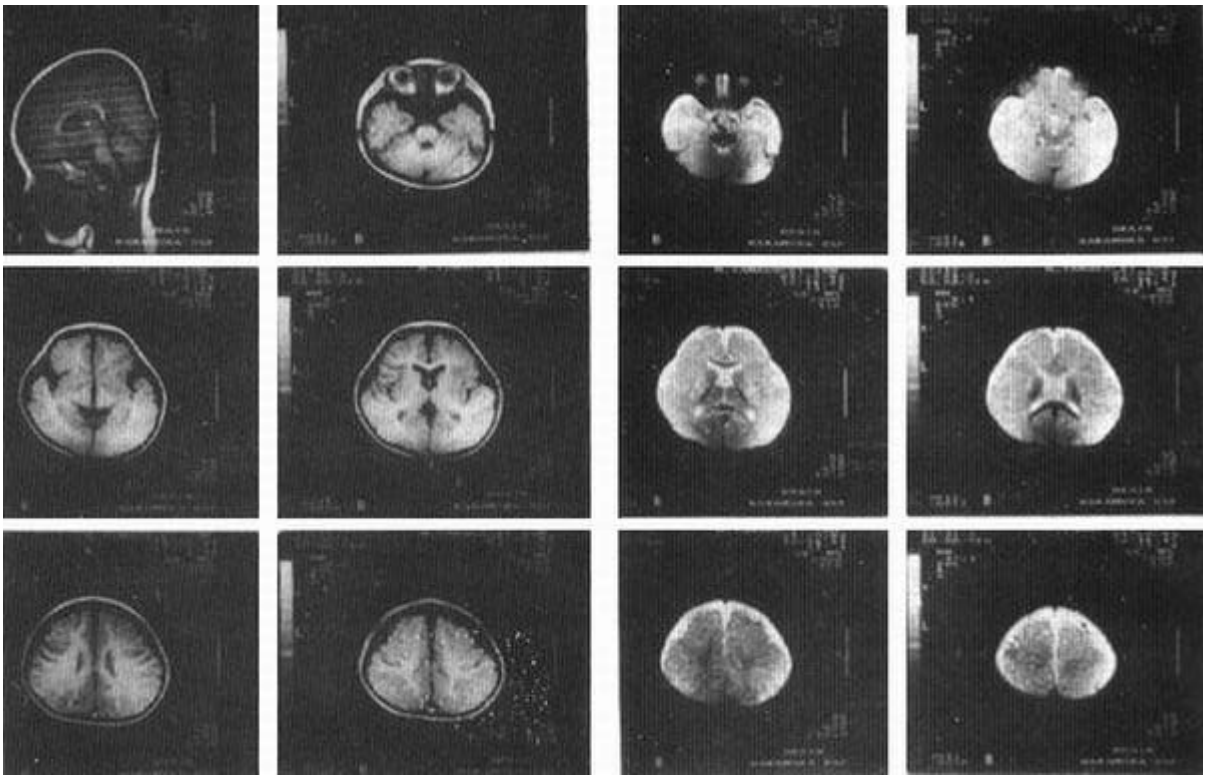

Figure 2: Transaxial T1(a) and T2(b) Weighted spin echo MR images with mild frontal atrophy and enlarged subdural space bliaterally, otherwise normal MR images.

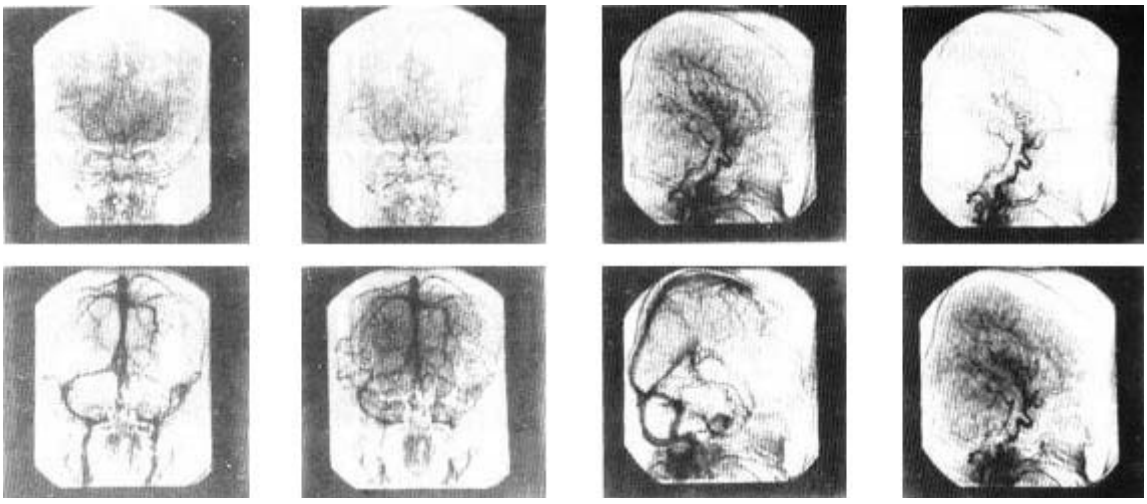

Figure 3: a, b: DSA with normal appearance.

equipped with a parallel hole collimator. Projection data were acquired over 64 angular views in a 64x64 matrix format, following i.v. injection of 2mCi IMP. Three hours later, delayed acquition was done. Both early and delayed data were analyzed and on early SPECT IMP brain perfusion imaging it was demonstrated that the whole of the right brain and left frontal lobe exhibited an apparent pattern of decreased tracer accumulation when compared to the other regions. On delayed images, almost complete filling of radioactivity to the areas described above was observed, indicating a reversible brain damage. Selected transaxial (early and delayed) images of the IMP SPECT stury are presented in (Figures 4a, b). The metabolic disorders were evaluated under the light of these results and metabolic work up revealed considerably high glycine level, 1117.56 nmol/ml (normal range: 18-331 nmol/ml). The diagnosis was confirmed as non-ketotic hyperglycinemia. After the treatment and dietary restriction, clinical improvement was observed. One month later, the follow up study with IMP SPECT showed an increase in tracer deposition in the regions previously displaying reduced tracer uptake.

DISCUSSION

In recent years, many diagnostic modalities such as CT, MR Imaging, SPECT have being extensively used in the field of Neurosurgery and neurology. It is well known that each of them image different pathophysiological processes (5-8,12,13). For instance, IMP-SPECT images reflect tissue perfusion (1), phereas techniques such as CT and MRI can provide information on tissue structures (8-10).

Although reports related to the evaluation of metabolic disease with these diagnostic modalities are quite limited, it was concluded that, CT could not allow a definitive diagnosis of metabolic disorders, but it is useful in the differential diagnosis and can be used to follow the progress of these disorders, looking for evidence of complications (3).

In the case of hyperglysinemia it was reported that CT scan might show diffuse cerebral edema, multicytic ensephalomalasia and cerebral atrophy (3). In our case, only a mild frontal atrophy, bilaterally was noticed both on CT and MR images. But only IMP SPECT revealed a large hypoactive area in the right hemisphere extending from the frontal to the pariatel and occipital lobe as well as in the left frontal lobe. It was the unique finding which could explain the clinical findings of the patient. Generally the underlying phenormena occurring with any hypoactivity on IMP SPECT images may be either a) decrease in blood flow, b) a functional deactivation, c) a metabolic impairment or a combination of these three parameters (2,4,11,14). Since DSA and MRI, could not reveal any blood flow abnormality, functional deactivation and/or metabolic impairment resulting in decreased uptake capability of the brain cells was thought as the main cause of the hypoactivity on IMP SPECT images.

The question of why MRI failed to show the abnormality seen on SPECT can be explained by the assumption that; the changing of the amount of H^+ ions in the lesions and surrounding area was inadequate to be detected by MRI. In another words this hypometabolism was not severe enough to produce perminant tissue damage or even to alter the water content of the area of lesion and surrounding tissue. As a matter of fact on delayed IMP SPECT images, to see the filling in to the areas seen as hypoactive on early images confirmed that this brain damage was reversible and rather mild.

So far, to our knowledge, this is the first report on comparison of CT, MR imaging and IMP SPECT in the patient with hyperglysinemia. IMP SPECT seemed to be more informative and sensitive investigative procedure than CT and MRI for the demonstration of even mild brain damage. It was also suggested that the lesion on IMP SPECT was due to the intracellular metabolic alteration rather than decreased perfusion and this suggestion might explain why MRI could not show any local lesion in this case.

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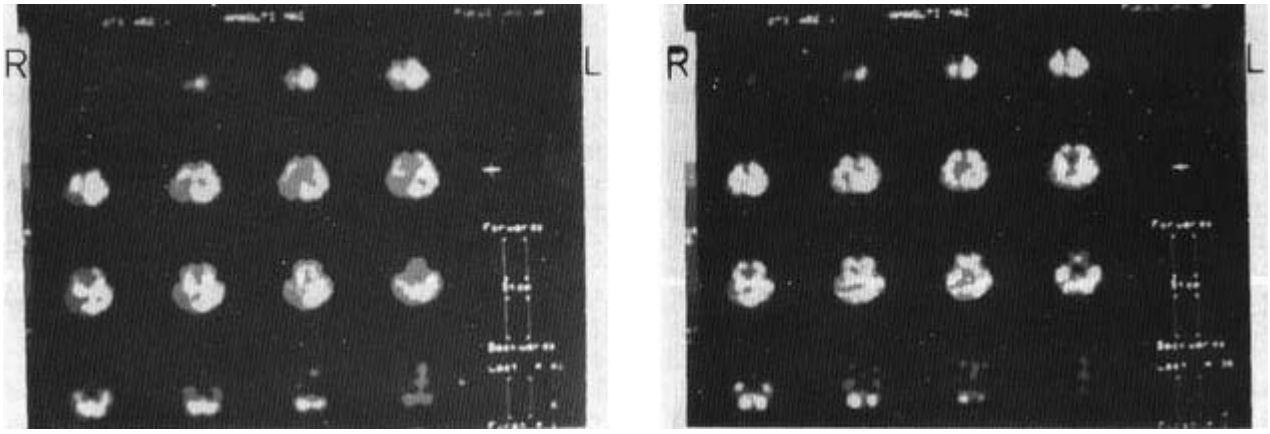

Figure 4: Transaxial early tomographic IMP images of the case (a) showing deceased tracer accumulation in the right hemisphere as well as in the left frontal lobe. Delayed images (b) revealing almost complete redistribution.

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