PERGAMON

Neuropsychologia 40 (2002) 245–252

NEUROPSYCHOLOGIA

www.elsevier.com/locate/neuropsychologia

Mania caused by a diencephalic lesion

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Received 21 February 2000; received in revised form 2 May 2001; accepted 23 May 2001

Abstract

We describe the case of a young male patient, SN, who suffered a MR-documented ischaemic lesion of both dorsomedial thalami and presented with a transient maniform syndrome. SN's neuropsychological, structural and functional imaging findings are compared with similar reported cases and are discussed in the framework of fronto-subcortical circuits and their proposed behavioural disorders. SN's mania was characterized by restlessness, mood elevation, a tendency for pleasurable activities, inflated self-esteem and loss of disease awareness. Other symptoms were sexual disinhibition, tactlessness, abnormal discourse, and reduced need for food and sleep. His neuropsychological assessment revealed an anterograde amnesia, and an impairment of frontal-executive functions. A SPECT-study showed diaschisis-related areas of hypoperfusion in both prefrontal regions which were interpreted as equivalents of SN's frontal-dysexecutive syndrome. In addition, there was a perfusion deficit in the right orbitofrontal cortex, which was taken as the imaging correlate of SN's secondary mania and personality disorder. These findings suggest that SN's mania and his other symptoms result from the twofold disruption of fronto-subcortical connections, namely of the right orbitofrontal loop which is concerned with mood regulation and socially appropriate behaviour, and of the dorsolateral prefrontal loop which mediates executive cognitive functions.

Keywords: Mania; Thalamus; Fronto-subcortical circuits; Orbitofrontal area; Dorsolateral prefrontal region

1. Introduction

The core behavioural abnormalities of the acute bilateral paramedian thalamic infarct (BPTI) comprise a state of somnolence and abulia accompanied by amnesia and a 'frontal brain' syndrome [38]. The disorder of vigilance often starts with a transient coma, followed by a gradually remitting stupor or hypersomnia during which patients present slow, apathetic and emotionally impoverished. During this state of 'psychic akinesia' spontaneous activity is decreased and patients require repeated and vigorous external stimulation during conversation or psychometric testing. With recovery of consciousness, behavioural and neuropsychological abnormalities become evident, mostly encompassing diencephalic amnesia [7,27,29,39], and frontal-executive dysfunction [10,33]. In addition to these standard findings, some case studies have described mood changes, such as cheerfulness, depression, lability with sudden switches from sadness to irritability, and outbursts of anger coupled with physical aggression [7,10,14,15,27]. Other forms of reported behavioural changes include sexual disinhibition and uncontrolled eating behaviour [8,15]. At present, no conclusive evidence exists as to exact origin of mood disorders following thalamic lesions. We report on a patient who suffered an acute manic episode due to a BPTI. The patient's findings are compared with similar, previously published cases in order to specify 'diencephalic mania' in greater detail, and an attempt is made to propose a neurological background hypothesis for the abnormalities of mood and cognitive behaviour following lesions of the diencephalon. Since it is known from experimental and clinical case studies that the frontal lobes are part of a complex neural network responsible for mood regulation and social behaviour [28], the approach adopted in this study will focus on the functional relationship

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0028-3932/01/$ - see front matter
PII: S0028-3932(01)00108-7

between the diencephalon and frontal brain areas, and on the consequences of a focal diencephalic lesion on this network.

2. Case description

SN, a 38-year-old previously healthy male commercial artist, was found deeply unconscious during a summer holiday in Turkey. During the next 48 h the patient gradually regained consciousness and was flown to his home country where he arrived awake and in good somatic condition, but logorrheic and profoundly amnestic. Apart from a complete vertical gaze paralysis and a slight dysarthria there were no other focal deficits. His history was free of drug abuse, previous neurological diseases, vascular risk factors or a recent trauma. No psychotic episodes were known in his family, and he had never gone through a period of mania or depression. MR-imaging depicted a fresh ischaemic lesion of both mediodorsal thalami on day 9 (Fig. 1). There were no ischaemic lesions in the frontal lobe. Since an echocardiogramm showed a patent foramen ovale, and a transcranial Doppler sonography revealed air bubbles after i.v.-ingestion, a paradoxical embolism causing a transient occlusion of the vertebral-basilary circulation was assumed, inducing the initial coma and an ischaemic BPTI, and the patient was put on i.v. heparin.

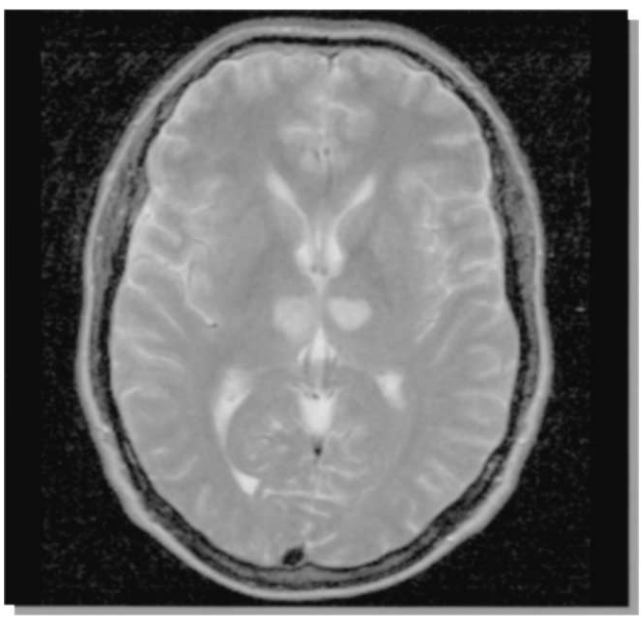

Fig. 1. MRT showing bilateral ischaemic lesion, including both dorsalmedial thalami; the lesion also includes parts of the intralaminar and anterior nuclei.

3. Behavioural findings

During the next 4 weeks SN's clinical picture was dominated by striking behavioural abnormalities. Different from other cases of the BPTI who present with hypersomnolence and apathy in the immediate period after their ictus, SN was continuously restless and agitated. He spent his time switching rapidly from one activity to another, writing letters, inventing and drawing new designs, portraying team members of the stroke unit, listing phone numbers from memory, or discussing plans for a wealth-growing future business. His mood was markedly elevated and his self-esteem inflated. He started long-winding discussions with every person he came across, producing tangential and often incoherent forms of speech. SN was easily distractable and had difficulties to stick to a topic, gave approximate answers (vorbeireden) and produced chains of 'klang-associations', puns and odd jokes. He also made many inappropriate or tactless comments and sexually suggestive remarks, behaved in a puerile and overfamiliar manner, had an exaggerated concept of his memory and cognitive abilities, and poor disease awareness. Though explicitly forbidden to him, he undertook spontaneous taxi excursions to the surroundings of the clinic. There were no signs of hallucinations or delusions, and he did not behave in an aggressive or irritated manner. His need for food and sleep was markedly decreased. It was soon noted that SN's memory was poor for the more recent time period, whereas he had no difficulties remembering distant, past events. His restlessness, agitation and extensive periods of wakefulness required low-dose treatment with a neuroleptic. Eight weeks after onset the majority of SN's maniform symptoms appeared markedly ameliorated and his medication could be discontinued. He had only limited memories of the manic episode.

4. Methods

Neuropsychological testing was performed on day 8 and during week 8 post disease onset. The assessment included standard tests such as the estimation of premorbid verbal intelligence [23], the MMSE [13], the Digit Span Test [43], the Trail Making Test [25], an oral test of mental arithmetics [20], and a test tapping visual organization [18]. Verbal and figural memory was assessed using an abbreviated version of the Münchner Gedächtnistest [19], a German equivalent of the California Verbal Learning Test [11], the Recognition Memory Test [42] and the Complex Figure Test [34]. SN's mnestic abilities and his tendency to produce provoked confabulations was further tested on a confabulation battery designed after Dalla Barba [9] tapping his personal (names and date of birth of family