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Short report

Focal lesion of the right cingulum: a case report in a child

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SUMMARY A boy with a focal lesion of the right cingulum, subjected to cingulectomy for the removal of a tumour, is the first case of an isolated unilateral cingulum lesion to be reported. The presenting symptoms consisted solely of serious behavioural abnormalities: lack of social restraint, heightened sexuality, bulimia and aggressiveness, all of which ceased after surgery. Neuropsychological tests, done before and after the operation, provided no evidence that the higher cognitive functions, including memory, were impaired.

The area of the cingulum has long been credited with neuropsychological functions, especially in regard to memory and behaviour. Its functional significance, however, is assumed rather than proven, and is derived from three types of investigations: (1) correlation of the anatomical and clinical findings in bilateral vascular or neoplastic lesions affecting this area,[1-3] (2) effects of stimulation during stereotaxic surgery,[4-7] and (3) clinical effects of bilateral cingulectomy for the treatment of severe mental illness or chronic cancer pain.[8-10]

Some authors have reported the consistent disappearance of behavioural abnormalities following bilateral cingulum lesions with appearance of apathy and, occasionally, transient confabulation in patients subjected to bilateral cingulectomy for aggressiveness.[8 9] In monkeys, a lesion or stimulation of the cingulum bilaterally causes transient behavioural changes, such as apathy and indifference to the group and to the environment.[11] Stimulation of the left cingulum only in, right-handed patients, is followed by a deterioration of short term verbal memory.[6] Stimulation of the right cingulum, on the other hand, is followed by no such deterioration. Apart from the stimulation studies, all these reports have been of bilateral lesions of the cingulum or lesions extending beyond it.

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We present a case of a child with a space-occupying lesion (astrocytoma grade 1) of the right cingulum that was surgically removed.

Case report

GS, a 10-year-old right-handed boy, had a 7 month history of seizures, that is sensations of alarm and groundless fear, autonomic phenomena (palpitations, pallor, profuse sweating, tremor, and mydriasis), simple compulsive movements of picking or touching, and more elaborate automatisms. At times these episodes were followed by loss of consciousness, urinary incontinence and tonic-clonic jerks, being complex partial seizures with secondary generalisation. Five months after the onset of the symptoms the child was given antiepileptic drugs (phenobarbitone with carbamazepine), which caused partial remission of the seizures. Shortly after, however, the patient developed an increasing behavioural disturbance with loss of social restraint, sexual polarisation, bulimia and aggressiveness. More or less simultaneously all his interpersonal relationships deteriorated progressively. It was at this point that he was admitted to the Istituto Neurologico, Milan.

On admission general and neurological examinations were normal, laboratory tests, skull and chest radiographs were all within normal limits. The visual fields could not be tested owing to lack of co-operation. During psychiatric examination the patient showed lack of restraint, hypomania, restlessness, exaggerated gesturing and talkativeness, heightened sexuality, which was displayed repeatedly in gesture and speech, and aggressiveness towards others. The obsession with sex was also manifested in a tendency

to exhibitionism with masturbation. The child made two attempts to escape from hospital. EEGs always showed a slow wave focus in the right frontal and frontoparietal regions and short bursts of bilateral sharp waves, synchronous in the rolandic and temporal regions of both hemispheres. Right carotid angiography was normal. A radioisotope brain scan showed a patch of increased uptake in the right frontal lobe and to the right of the midline at the level of the genu of the corpus callosum. Computed tomography (CT) showed a space-occupying lesion in the right frontal region just opposite the genu of the corpus callosum, which indented the frontal horn of the right ventricle (figure). Craniotomy was performed; a 2 cm long and 2 cm deep transverse incision of the cortex (F1 convolution) revealed a well-defined greyish nodule with a diameter of 1·5–2 cm located in the gyrus cinguli, immediately in front of the frontal horn of the right ventricle.

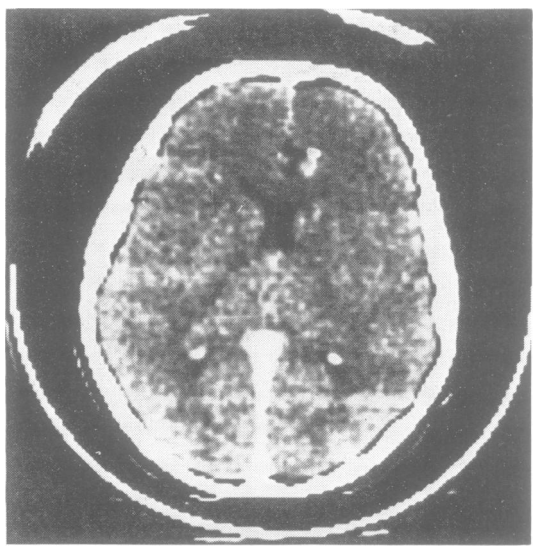

Figure *Post contrast CT scan. The lesion is within the right cingulate gyrus and impinges upon the genu of the corpus callosum, partially obliterating the right frontal horn. Posterior to calcification, there is a small area of enhancement.*

A portion of brain tissue in the cingulum region was removed together with the tumour. Histological examination showed this to be a grade 1 astrocytoma. The postoperative course was uneventful. Fifteen days after the operation neurological examination was normal. Psychiatric examination showed reduced impulsiveness and aggressiveness. The patient's thinking and talking were no longer concentrated on sex, but he was emotional and unduly alarmed by trivial but invasive medical manoeuvres.

Follow-up at 3 and 12 months showed that his epileptic fits had stopped, and the aggressiveness, lack of restraint and fixation on erotic themes had ceased. EEGs were done 15 days, 3 and 10 months after the operation; the last two EEGs showed only symmetrical minimal theta wave anomalies. The CT scan done 20 days after the operation confirmed complete removal of the tumour in the right cingulum area and enlargement of the right ventricle. In view of the peculiarity of the lesion site, we administered projective tests (Rorschach and TAT) and a series of neuropsychological tests which are summarised in the table.

Table *Summary of intelligence and memory tests*

Intelligence tests	*Preoperative score*	*Postoperative score*
Progressive matrices 47	19	26
WISC:		
Full scale IQ	85	109
Verbal Scale IQ	94	96
Performance Scale IQ	78	122
Memory tests	*Form I*	*Form II*
Digit span forward	5	6*
Digit span reversal	2	3*
Digit span with supraspan	Non-criterion	Non-criterion
10 Words learning	7	8*
Logical memory		
im	15	10†
−1 h	10	8†
Block tapping test (Corsi)	4	4
Supraspan tapping test	Criterion: 10^	Criterion: 6^*
12 Shapes learning	7	8*
Memory for rhythms	7	7
Motor memory	7/10	9/10
Memory for faces	10/10	8/10†
Memory for noises	10/10	8/10†
Rey complex figure reproduction	9·5	Milner complex figure 7†

Binomial test (N $12 \times 6 = 613$ p=NS).

The variations (improvement *, deterioration † in memory tests scores are not significant (Binomial test).
The mean scores of these tests were obtained at the Neuropsycologisches Laboratorium of Zurich Neurologische Universitatsklinik (dr E Perret) in 8–12 year old children. The mean scores represent the means for within-case comparisons.

Conclusions

The findings before cingulectomy may be summarised as follows: (a) behavioural abnormalities: lack of restraint, aggressiveness, undue concentration on sex and bulimia with pathological equivalents in the responses to the projective tests; (b) insignificant memory deficits both for verbal and for non-verbal material; (c) low scores in the PM 47 test, on the performance scale of the WISC and in reproducing of Rey's Complex Figure from memory.

The findings after operation were as follows: (a) cessation of behavioural abnormalities with cessation of the more pathological percepts in the projective tests, especially in the Rorschach test; (b) no significant changes in the memory functions; (c) higher scores in the PM 47 test (inexplicable in terms of benefit of retesting),[12] and even better scores on the performance scale of the WISC but no improvement in reproducing the Milner Complex Figure Form II from memory.

The behavioural abnormalities before the operation could be interpreted as a chronic functional disturbance caused by the epileptogenic focus round the tumour, maybe also disinhibiting the temporal regions, for the patient suffered from complex partial seizures and exhibited an EEG-proven epileptogenic focus. This hypothesis would account for the cessation of the seizures, of the EEG focus and of the behavioural abnormalities after cingulectomy. Thus removal of the epileptogenic focus around the tumour, rather than cingulectomy *per se*, was probably what eliminated the behavioural abnormalities.

Finally, in regard to memory functions and their correlation with the cingulum areas, the analysis of our case contributes something to this long debated and still unresolved problem,[13] in that it shows that unilateral cingulectomy *per se* does not appreciably affect memory.

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