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Case Report

Possible common neurological breakdowns for alexithymia and humour appreciation deficit: A case study

Panayiotis Patrikelis[a,*], George Konstantakopoulos[b,c], Giuliana Lucci[d], Vasileios K. Katsaros[a,e], Damianos E. Sakas[a], Stylianos Gatzonis[a], Giorgos Stranjalis[a]

[a] Department of Neurosurgery, School of Medicine, Evangelismos Hospital, University of Athens, Greece
[b] First Department of Psychiatry, School of Medicine, Eginition Hospital, University of Athens, Greece
[c] Section of Cognitive Neuropsychiatry, Department of Psychosis Studies, Institute of Psychiatry, Psychology and Neuroscience, King's College London, UK
[d] Unit of Neuropsychology, IRCCS Santa Lucia Foundation, Rome, Italy
[e] Department of MRI, General Anti-Cancer and Oncological Hospital of Athens "St. Savvas", Athens, Greece

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ABSTRACT

This brief work is an attempt to point to the possible common neurological breakdowns in giving rise to alexithymia, and impaired appreciation of humour. In particular, we present the case of a patient who lost the ability to enjoy humour after the surgical removal of a frontal groove meningioma, although he was still able to detect it, while at the same time was diagnosed with organic alexithymia. Our results indicate that problems in the affective appreciation of humour and in emotionalizing (alexithymic symptoms) may be the result of damage to the ventral-rostral portions of the ACG/mPFC, which prevent the patient from assessing the salience of emotion and motivational information, and generating emotional reactions; as a result he has trouble experiencing emotions, knowing how he and others feel, and enjoy humour.

1. Introduction

People who have sustained brain injury may manifest symptoms of alexithymia which might be referred to as 'organic alexithymia'. Organic cases of alexithymia which symptoms firstly appear as a result of a brain lesion and in the absence of other types of psychopathology are not well understood and largely neglected. It is also referred to as 'secondary alexithymia' and is often more resistant to treatment, while it is not associated with a specific premorbid personality and is associated with cognitive impairment. The lack of the sense of humour is also an important aspect of alexithymia although widely underestimated and far from being well understood.

2. Case report

In the following, we present a patient who lost the ability to enjoy humour after the surgical removal of a frontal groove meningioma, although he was still able to detect it, while at the same time was diagnosed with organic alexithymia.

Our patient is a 47-years-old right-handed man (Mr. F.J.), teacher of physics in high school, who received surgery for a giant frontal groove meningioma with a maximum diameter of 7 cm, occupying the frontal cortex bilaterally. In particular, there was evidence of an extra-axial space occupying lesion with morphological characteristics of a meningioma causing posterior and downward displacement of the optic chiasm and the pituitary gland, as well as compression on the frontal gyri and the frontal horns of the lateral ventricles with slight peritumoral edema, especially on the right of the mass (Fig. 1A). Preoperative tumour embolization was considered necessary, however it was considered as impossible. A week later the tumour was totally resected. After surgery the patient presented irritation and confusion, as well as paranoid ideation and therefore received antipsychotic medication (haloperidol 2 mg/d). About five days later, the patient ameliorated considerably. The follow-up MRI scan 1 year after the operation showed midline bilateral frontal changes characterised by gliotic and porencephalic areas, especially in medial prefrontal cortex (mPFC) and anterior cingulate gyrus (ACG), as well as the cingulum and anterior part of corpus callosum (Fig. 1B). Fourteen months after surgery, when the patient came to our observation, his neurological condition was

* Corresponding author at: 45-47 Ipsilantou Str., 10676 Athens, Greece.
E-mail address: ppatrik@cc.uoi.gr (P. Patrikelis).

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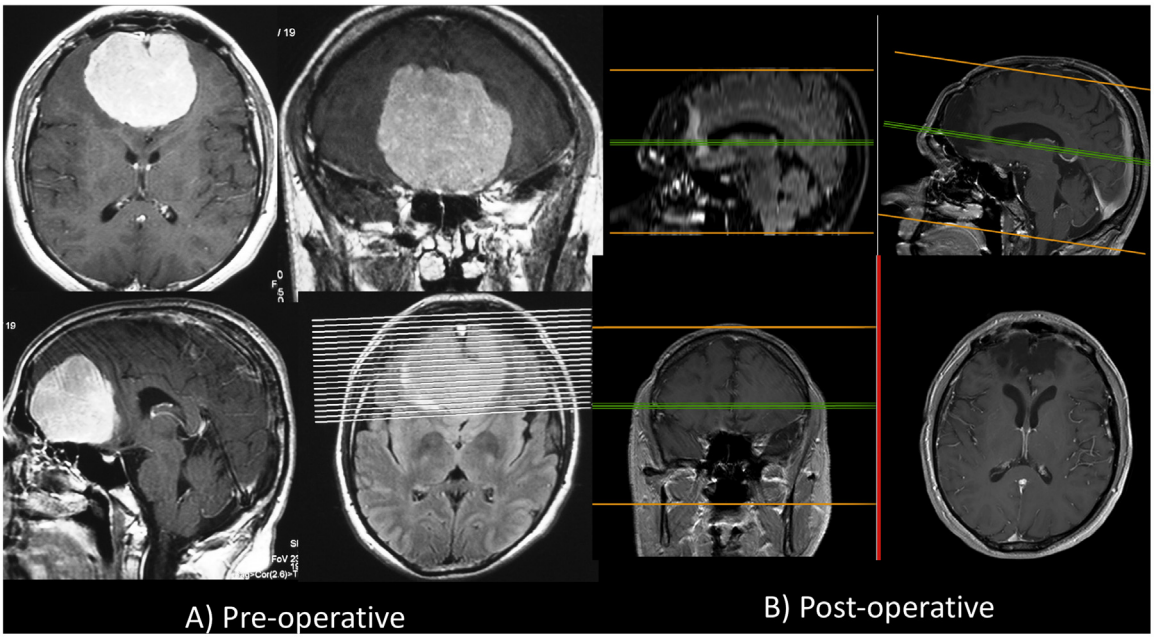

Fig. 1. (A) Pre-operative MRI: 3 planes post-Gd T1-weighted images, demonstrating intense enhancement of the meningioma and axial FLAIR image (lower right), showing peritumoral edema. (B) Post-operative MRI: Sagittal FLAIR image (upper left) and post-Gd T1-weighted images in 3 planes, confirming the total excision of the mass and showing findings consistent with porencephaly and gliosis of the medial prefrontal cortex, anterior cingulate gyrus, as well as the cingulum and anterior part of corpus callosum.

normal, as confirmed by an objective neurological examination, with exception to a persisting anosmia.

At that time F.J. claimed that he had lost the ability to appreciate the sense of the humour *(the affective experience of mirth),* while he reported being still able to detect it *(getting the joke).* This loss was especially disrupting, since he used to be a person with an excellent sense of humour in the past.

Psychiatric examination confirmed that psychotic symptoms had remitted completely and revealed no mood or anxiety disorder. However, F.J. reported that he could not easily identify the feelings he was experiencing and he frequently failed to share others' feelings, which led to difficulties in social interactions. He had not recognised these impairments until his partner pointed out that he became emotionally distant and socially withdrawn. Both the patient and his partner mentioned that 'his character changed' after surgery. However, F.J's wife constant emotional cueing and coaching proved to be invaluable, since his decreased emotional awareness and intention attribution biases *(see below)* rendered interactions sometimes problematic, particularly those with colleagues at work. The psychiatric assessment concluded that the current diagnostic criteria for alexithymia [1] were fulfilled. Moreover, the Toronto Alexithymia Scale-20 total score was 72, which is above the threshold of ≥61 for clinically important alexithymia. F.J.'s neuropsychological evaluation showed a decrease in his verbal abilities (VIQ) and problems in mental flexibility, intention attribution problems (Faux Pas Recognition Test), and humour and metaphor comprehension (see Table 1). The above difficulties probably reflect a declining trend from patient's premorbid level of cognitive functioning, but not a general mental decline or a broader functional deficit.

3. Discussion

It has been assumed that a pool of anterior brain structures (i.e. prefrontal and anterior cingulate cortex (ACC), insula (INS), amygdala, corpus callosum and anterior commissure) plays an important role in emotional processing and expression of alexithymia [2]. Berthoz and colleagues [3] sustained that alexithymia, as a deficit of higher order appraisal of emotional stimuli, may be linked to the ACC and the mediofrontal cortex (mFC) malfunction, while the amygdala, the hippocampal formation and the hypothalamus, which process lower order emotional aspects (i.e., the perceptual information) of stimuli result intact. They concluded that alexithymia operates exacerbating personality traits, and it usually plays a role in affect regulation associated with circumscribed valence-dependent differences in the activity of the ACC and the mFC during emotional stimuli processing [3].

The most prominent neurophysiological theory of acquired alexithymia [4] links alexithymia to *functional commissurotomy*, assuming that abnormal interhemispheric communication reduces coordination and integration of specialized functions and determines an abnormal state of inhibition.

Impaired humour appreciation may be related to deficits in processing positive or happy stimuli which could occur in alexithymia. It is known that deficits in processing of or responsivity to negative stimuli can also be observed in alexithymia. Humour processing entails many cognitive aspects, such as the set-up of the joke, associated with temporal lobe (i.e., temporal pole and anterior superior temporal sulcus) activation [5]; humour comprehension and detection/resolution of incongruity, involving the inferior frontal gyrus [6] and regions around the temporal parietal junction [7]; the attribution of mental states to the characters of the joke, related to the activation of the anterior medial prefrontal cortex (amPFC) [8]. The affective experience components of humour processing are related to the medial ventral prefrontal cortex (mvPFC), subcortical nuclei (nucleus accumbens), bilateral INS and left amygdala functioning [6,5].

In summary, literature points out the crucial role played by cortical midline structures (in particular, mPFC and ACG), INS and amygdala in both alexithymia and humour appreciation deficit, in agreement with a recent theory assuming the critical role of anterior cortical midline structures in emotional feeling [9].