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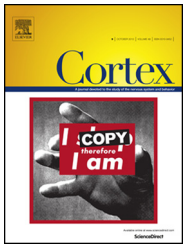

Research report

State of belief, subjective certainty and bliss as a product of cortical dysfunction

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ABSTRACT

Introduction: Ecstatic seizures are focal epileptic seizures which are fascinating from a phenotypical point of view as they include intense positive affect, feelings of heightened self-awareness and enhanced well-being. They have been previously suggested to arise in the anterior insular cortex, although strong arguments are still lacking.
Methods: We describe the cases of two new patients with ecstatic seizures. Their evaluation included a careful history, encouraging the patient to provide significant details about their ictal symptoms in order to better understand the origin of the sense of bliss and support the hypothesis of an insular involvement according to the current stage of knowledge. Ictal electroencephalographic and blood flow studies complemented these data in one patient.
Results: The comprehensive description of the ictal ecstatic symptoms by the two patients has brought out an unfamiliar sense of absence of doubt which was at the basis of a feeling of meaningfulness and certainty. The ictal single-photon emission computed tomography (SPECT) showed an increased blood flow maximal at the junction of the right dorsal mid-insula and the central operculum.
Conclusions: The unveiling of an ictal sense of certainty during ecstatic seizures might imply, in the light of current knowledge, a defect in the system processing prediction errors within the framework of generalized predictive coding mechanisms of the brain. Accumulative evidence has recently highlighted a crucial role of the anterior insular cortex in this system, particularly in the detection of mismatch/conflict between prediction state and outcome. Abnormal activity related to epileptic seizure in a structure prevents its normal activity: in the anterior insula, it could prevent the detection of prediction errors, and thereby prevent the feeling of ambiguity (and the associated negative emotional component), leading to a blissful state which could be close to the deeper states of meditation.

1. Introduction

During recent years there has been growing evidence for the role of the insula in many important cognitive processes and particularly in dealing with uncertain/ambiguous information. It was first recognized as the main cerebral structure behind our sense of global interoception, generated at every moment via the processing of various sensory external and

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internal stimuli (Craig, 2009). An integrative model of the role of the anterior insula in feelings and processing uncertainty in the context of decision making has been proposed (Preuschoff et al., 2008; Singer et al., 2009). The insular cortex seems indeed to integrate external sensory and internal physiological signals with computations about their uncertainty. This integration is expressed as a dominant feeling state that modulates social and motivational behavior in conjunction with bodily homeostasis. As we are going to show in this paper, the role played by the anterior insula in processing uncertainty could be of major importance to account for the "ecstatic" symptom.

The first ecstatic epileptic seizures that we can recognize throughout the literature are those of Dostoevsky, in his correspondence and also attributed to some of the characters of his novels, as the Prince Mychkine in "The Idiot". Ecstatic seizures consist of a feeling of bliss, associated with a sense of high self-awareness and a physical sensation of global well-being (without any sexual feeling). The ecstatic seizures have been suggested to originate in the insular cortex (Picard and Craig, 2009) because (i) the anterior insular cortex is the best candidate region that may explain concomitant feelings of heightened self-awareness (Craig, 2009; Modinos et al., 2009; Critchley et al., 2004), enhanced well-being (Craig, 2003) and intense positive affect; and (ii) ictal single-photon emission computed tomography (SPECT) performed in two patients have shown increased blood flow in the anterior insula (Picard and Craig, 2009; Landtblom et al., 2011).

The present study reports the phenomenologic description brought by two new patients with a high power of introspection, intelligence, and level of vocabulary, which helps to dissect the cognitive process leading to the ecstatic feeling.

2. Patients and results

2.1. *Patient 1*

A 17-year-old Swiss apprentice farmer suffered from epileptic seizures from the age of 15 years.

He experienced an extremely pleasurable state of consciousness during his seizures. He felt a deepening awareness of the situation or conversation going on around him, a sudden clarity. It was as if he clearly understood everything, especially if he happened to be in the midst of a discussion with several people. He grasped it all simultaneously. Things suddenly seemed self-evident, almost predictable (yet without the feeling of knowing the future).

While he knew where he was and what he was doing, he did not want to interrupt the experience by talking. At the same time, very often he felt a taste, pleasant but nothing more (gustatory hallucinations). He could not compare this taste with any taste he knew. During the seizure, he could not "be located in time". It was as if time stopped. For him, these seizures lasted at least 40 sec while his family members could say it lasted 1–3 sec. These seizures were all triggered by a pleasant context. It could be an interesting discussion with several people with whom he liked to talk. In this case, the significance of all parts of the discussion suddenly became completely clear, as if a sudden incredibly clear understanding and meaningfulness of the whole discussion was offered to him. Other triggering factors were nice things that he could see, hear or think about. It could be when he saw "a tractor with the harvest, nice photos, a nice color, a flower, a nice landscape, a bird singing, grazing animals, branches that move with the wind, a beautiful woman", or on the occasion of a kiss, a caress, a nice thought about someone, a hope….

At the beginning of his epilepsy history, he suffered from more severe focal seizures, without any clear triggering factor, starting with a very strange bad taste, unsimilar to any existing taste, a vestibular illusion and hypersudation, followed by a loss of consciousness with oral automatisms (chewing movements) and then a tonic seizure of the face and upper limbs, with prolonged post-ictal confusion.

Magnetic resonance imaging (MRI) showed a tumor suspicious of a ganglioglioma in the right temporal pole beside an arachnoidal cyst (Fig. 1A). An 80 sec-seizure with ecstatic symptoms (the patient seemed struck by a sudden thought) followed by a loss of consciousness with left hemifacial dystonia and then oro-alimentary automatisms was recorded. Ictal electroencephalogram (EEG) showed rhythmic 12 Hz activity in the right anterior temporal area, with a diffusion in the homolateral frontal region, followed by delta slow waves in the bilateral frontotemporal regions and then a right anterior temporal flattening at the end of the seizure. A moderate tachycardia was observed from the beginning of the clinical

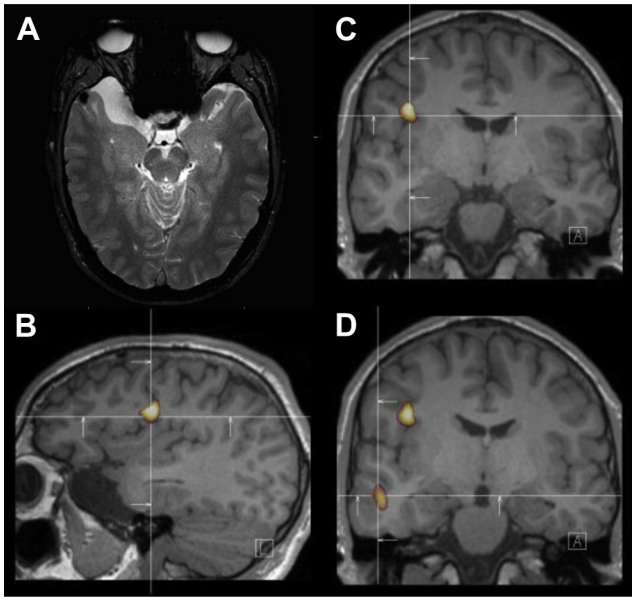

Fig. 1 – Cerebral MR and ictal SPECT images of patient 1. (A) Gradient echo T2* axial MR image showing a small round (hypointense) tumor in the right temporal pole and a neighboring arachnoid cyst. (B–D) Ictal/interictal technetium-99m HMPAO (^{99m}Tc-HMPAO) SPECT subtraction using BRASS analysis program. The software allows automatic fitting of brain perfusion scans and subtraction; (B) sagittal image, (C–D) coronal images, showing an increased blood flow maximal at the junction of the right dorsal mid-insula and the central operculum. (D) A cluster of increased blood flow was observed in addition within the right temporal pole, close to the tumor.