

Poststroke Emotionalism

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Introduction

Poststroke emotionalism (PSE) has a high prevalence (15-35%) [1,2]. Though available data is limited, it suggests an increase which parallels the time elapsed since stroke onset, at least during the first three months [3].

Emotionalism might be one main symptom within a variety of neurological disorders of vascular, traumatic, degenerative, neoplastic or inflammatory origin. PSE is also frequent in patients with psychotic and primary mood and affective disorders as well as in patients with depression due to other medical or neurological conditions. Hence, despite different pathogenic mechanisms, PSE appears to be related to a similar topographic pattern of lesion distribution or dysfunctions. However, although in this context PSE should be considered a disorder which belongs to the field of clinical neurology, its relationship with affective disorders and emotional control processes in general makes it a privileged field of study in neuropsychiatry, behavioural neurology and neurosciences.

PSE is an increase in the frequency of crying (shedding tears, sobbing) or laughing episodes in comparison to the patient's condition before the stroke event. With PSE the onset of such episodes occurs with little or no warning, the patient feels that emotional expression goes beyond the normal control, and he might cry or laugh in social contexts when she or he would not previously behaved as such [1]. Finally, crying and laughing are not only more frequent in patients with PSE than in healthy people but are also more intense or excessive than simply a few tears or smiles. While most patients with PSE exhibit either crying or laughing behaviour, relatively few manifest both behaviours. Several alternative definitions are employed for PSE such as spasmodic, forced, involuntary, inappropriate, and uncontrolled or dissociate laughter and crying, pseudobulbar affect, emotional incontinence and involuntary emotional expression disorder. The different abovementioned denominations serve to underline the character of the emotional expression (crying or laughter) which manifests in patients merely beyond voluntary control. The reflex nature of this behaviour reminds in some way to other automatic behaviours in neurological disorders (spasms and spasticity, dystonia, psychomotor epilepsy, catastrophic reactions, and environmental dependent behaviours).

Nevertheless a parallelism could be traced with situations that are frequent for some healthy individuals. For example, some people declare being not able to control themselves in preventing tears or crying at movies even when this behaviour is adverted as embarrassing toward the one nearby seated. Also in healthy people, crying and laughing tend to maintaining themselves once they have started. Every one experienced that these acts lack an "on-off" switch. Furthermore, crying and laughing are sometimes seen as contagious, and the laughter of one person can itself provoke laughter or crying from others, a situation that can escape to efforts of any voluntary self-control. Finally, loss of control on laughing and crying is a physiological condition of the human behaviour itself.

Based on clinical observations, Poeck et al. proposed that PSE could be probably differentiated in two further conditions: pathological laughter and crying (PLC) (or pseudobulbar affect [PBA]) and

emotional lability (EL) [4,5]. However, this dichotomy appears to be in some way simplistic even on clinical grounds, as PLC and EL should be better considered two conditions along a continuum.

PLC differs from EL on the basis of some clinical features. Crying and laughing have a more reflex quality in PLC than EL, which is suggested by their occurrence in PLC with little or no latency after irrelevant stimuli and by the frequent absence of congruence between the emotional expression and the internal affect. For instance, PLC can occur during relatively unemotional situations such as normal conversations, physical exercise, medical examinations or even be triggered by ocular movements or unexpected sounds (i.e. the telephone ringing). In summary, PLC presents the following characteristics: 1) the behaviour is triggered by unspecific stimuli; 2) lack of relationship between the emotional expression and affective changes; 3) the absence of a corresponding change in mood during or lasting beyond the actual laughing and crying; 4) the difficulty in controlling his own facial expression during laughing and crying [5].

Concerning EL, crying and laughter are generally provoked by stimuli that have emotional significance (e.g. hearing "bad news", seeing a beloved one) and, although the behaviour occurs abruptly and is experienced as uncontrollable, the patient feels congruent emotions (joy or pleasure in the case of laughing and sadness or discomfort in the case of crying).

These general definitions allow formulating the hypothesis that PLC and EL might result from different neural mechanisms, i.e., for PLC, a defective control of the motor acts of crying and laughing, and, for EL, a defective control on the emotional experience. However, these assumptions remain still speculative since they have not yet been subjected to investigations in stroke patients, which are specific to that dichotomy. Furthermore the distinction of PLC from EL only on the basis on the congruence between trigger stimuli and internal feelings is often clinically difficult and impractical.

It is also unknown whether neural systems of crying and laughter are the same, to what extent they overlap and whether they are subjected to the control of similar cognitive or motor processes.

Anatomical Correlates of Emotionalism

In 1924, S.A.K. Wilson formulated a general theory on the neural correlates of emotionalism in neurological disorders [6]. This theory postulates the existence of two cortical systems connected by

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the corticospinal tracts to a hypothetical “crying-laughing” facial-respiratory center in the brainstem, responsible for the motor commands of crying and laughter. The first system, located in the frontal lobes and in the motor cortex, exercises a volitional control, whereas the second, presumably connected to the limbic system, processes the emotional valence of the external stimuli and allows laughing and crying emerge to involuntarily. According to this theory, the emergence of abnormal laughter and crying is due to the imbalance between the two systems and, for this reason, may manifest even after unilateral lesions. Somewhat later, Papez hypothesized that supranuclear pathways, including those from the limbic system, mediate emotional expressions and synapse in the reticular core of the brainstem [7]. This hypothesis frames into later theories, imputing the tegmentum near the periaqueductal gray matter to contain the integrative mechanism for emotional expression.

However, despite their appeal, these theories were only vaguely formulated and could thus be neither included in cognitive models of emotional control nor yet substantiated in terms of brain functional systems.

For PLC the involvement of the corticospinal tracts is strongly suggested by case reports where the disorder manifested itself after uni- or bilateral ischemic or hemorrhagic stroke in the basis pontis [8-10]. PLC has been reported as a presenting symptom of subtentorial tumors compressing the pons or the midbrain [11,12]. In some of those cases PLC completely disappeared after surgical resection of the tumor [13].

The role of the pontine pathways is also suggested by the insurgence of the “Fou rire prodromique” (an uncontrollable crisis of laughing that shortly anticipates a stroke [14]) before ischemic or hemorrhagic strokes occur, which are generally located in the basis pontis [15,16].

However, the role of other cortical and subcortical structures in the pathogenesis of laughing and crying is suggested by “gelastic” (laughing) [17] and “dacrystic” (crying) epileptic crisis [18] in patients with hypothalamic hamartomas, or tumors or other lesions in the temporal [19,20] or mesial frontal lobe [21], and finally, by the effect of in situ electrical stimulation of cingulate and basal temporal cortex [22] and subthalamic nucleus [23,24].

An alternative hypothesis, based on a single case report of a patient with multiple brainstem and cerebellar lesions [25], attributed to the cerebellum the role of modulating and adjusting laughter and crying behaviors to the cognitive, emotional and situational values of triggering stimuli.

However, a specific linkage between EL and lesion location has not yet been demonstrated in a larger cohort of stroke patients. Anterior cortical or left front allenticular-capsular lesions, particularly those involving the dorsal globus pallidus, temporal lobe and thalamus have been indicated as possible correlates in clinical studies that compared CT or MRI findings between groups of stroke patients with and without EL [1,2,26-28].

These findings have to be considered cautiously due to some methodological limitations, specifically because of the different criteria employed for establishing PSE.

In our opinion, even after detailed questioning, for most patients with PSE it is impossible to determine exactly whether the disturbance occurs at the level of the emotional processing or emotional expression or most likely, on both levels. Thus, new behavioural paradigms should be specifically designed.

Neurobehavioral data aiming to establish a link between PSE,

cognitive deficits [27] and other mood (anxiety and depression) and emotional disorders (i.e. catastrophic reactions, empathy or theory of mind deficits) are limited for stroke patients.

In healthy individuals functional neuroimaging studies (with PET and fMRI) showed that paradigms of sadness or happiness induction (potential triggers of laughing and crying) activate a great number of cortical regions. The correlations are more robust for sadness and consistent with the anterior cingulate cortex and insula [29]. Human laughing and crying activate insula and amygdala regions [30] while imitation or suppression of happiness or sadness activates insula (for the emotional processing) and prefrontal or dorsolateral regions (for control processes) [31].

Although the anterior cingulate cortex and specific limbic areas such as amygdala and insula are only rarely and selectively damaged by ischemic stroke, their reciprocal projections to the frontal lobes, limbic striatum, and other paralimbic areas are very often involved in large strokes. Given the central role of prefrontal-subcortical-amygdaloid systems in emotional regulation [32,33], it is highly conceivable that the loss of control over crying and laughing is intimately related to the dysfunction of those systems.

Neurochemical Findings, Psychiatric Aspects and Drug Response

A neurochemical hypothesis postulates that PSE is the consequence of a dysfunction of serotonergic neurotransmission. This hypothesis is supported by some PET- mapping results of serotonin receptors in patients with PSE [34] and by the rapid (few days) abortive effect on crying and laughing by selective serotonin reuptake inhibitors (SSRIs) such as sertraline [35], paroxetine [36], fluoxetine [37,38] and citalopram [39].

Furthermore, the brainstem raphe nuclei (the main site of serotonin synthesis) give rise to serotonergic projections to the limbic forebrain, and serotonergic receptors are widespread in many brain regions, especially in paralimbic areas. This wide distribution of serotonergic fibers and receptors makes them vulnerable to every stroke and could explain the heterogeneity of lesion location attributed to PSE.

Moreover, polymorphisms of serotonin transporter (5-HTT) gene have been found to be a susceptibility factor for PSE in an East Asian population [40]. However, PSE responds to tricyclic antidepressants, dopaminergic and other non-SSRI drugs [41], although it cannot be excluded that this effect could be still mediated indirectly by the serotonergic system.

From a psychological point of view, PSE has been considered as one manifestation of a more general disorder of emotional control, thus being related, for example, to the post-traumatic stress disorder. Stroke victims can lively remember the occurrence of a stroke as a traumatic event that severely disrupted their existences. From this perspective, the process of coping with the disease produces irritability, intrusive thoughts and memories that could interfere with emotional regulation [42]. Furthermore, PSE causes distress, embarrassment, social avoidance, and impaired quality of social interactions. Other people may perceive sudden crying or laughing as a sign of dementia or stupidity.

PSE has been found as a risk factor for developing poststroke depression (PSD) within the first year after stroke onset [43] and both PSE and PSD favourably respond to SSRI drugs [26]. These findings suggest a further common pathogenic mechanism for both disorders.

PSE, as well as PSD, might be factors of negative functional outcome and reduced quality of life for stroke patients [44].

Response to SSRI does not appear to be the result of a simple antidepressant action since recovery also occurs in people without a depressive disorder soon after starting a low dose of the drug [45]. In a few cases, a pharmacological treatment transformed crying into laughter [46].

Although antidepressants (specifically SSRIs) appear clinically efficacious, results obtained from meta-analysis do not provide specific recommendations for a specific class of drugs [47]. It would be also of clinical interest to assess in future studies whether the response to these drugs is sustained after their withdrawal.

Future Directions and Conclusions

Emotional disturbances have occupied clinicians from the last century to nowadays since they can occur in a variety of neurological conditions. PSE probably constitutes a heterogenic group of emotional disorders that has high prevalence, is possibly related to a serotonergic dysfunction but still lack specific neural correlates.

Available data suggest a diffuse neural network which is involved in emotional perception cognitive and control processes as well as in motor acts. Neurologically oriented theories of emotion have evolved from the phenomenological examination of individual patients with affective changes after cerebral damage. Together with initial findings from animal studies on the basis of behaviouristic paradigms some theories evolved around the turn of the last century proposing some mechanisms of emotion, based on evolutionary assumptions of the wiring of some human brain systems. Meanwhile, appraisal theorists emphasized the role of the relationship between an organism's goals, plans and coping strategies rather than simpler stimulus-response associations. Unfortunately, there have been only sparse attempts to integrate neurological data together with functional neuroanatomical findings in order to develop a neurocognitive theory of emotional processing which might explain pertinent clinical observations. Thus, while on one hand clinical studies describe disrupted behaviour on the basis of distinct brain damage, on the other hand psychological approaches are aimed to explain how organisms acquire knowledge of the value of stimuli in the world. However, only few approaches account to converge into a unified theory of emotional processing.

Only behavioural, neuropsychological, neurofunctional imaging and neurochemical studies, together with neurogenetic approaches on very large cohorts of patients with emotionalism after stroke might offer an unique opportunity to investigate emotional regulation and expression and might hence lead to a deeper understanding on the complex mechanisms of emotional processing in health and disease.

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