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Emotional Dysregulation in Psychogenic Voice Loss

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Sigmund Freud first proposed that emotional distress can produce symptoms of pseudoneurological dysfunction [1]. Thus, the idea that strong aversive emotions can disrupt the brain's functional integrity dates back to early hysterical conversion concepts. Intriguingly, conversion disorder still constitutes one of the few psychiatric conditions where diagnostic classification systems take account of etiology, specifying emotional distress as an initiating factor [2]. Patients typically present neurological symptoms of motor or sensory dysfunction in the absence of an underlying organic condition. Despite the long-standing interest in conversion disorders, neuroscience has only recently begun to explore its neurobiological signature [3].

Current conceptualizations hypothesize that conversion symptoms may result from disruptions of brain networks engaged in processing emotions. Animal models and human imaging studies suggest that chronic exposure to high and uncontrollable stress is associated with profound adaptation of neural responses in the amygdala and prefrontal cortex [4]. These regions are essential for emotion perception and regulation, respectively, and interact as core amygdala-prefrontal circuit within the emotion-processing networks [5]. Indeed, accumulating evidence from neuroimaging research in patients with stress-related psychiatric disorders, particularly posttraumatic stress disorder (PTSD), indicates that pathological disruptions in this amygdala-prefrontal circuit might represent a common denominator underlying the detrimental effects of stress on brain functioning [6, 7].

Here, we aimed to examine the integrity of the amygdala-prefrontal circuit in patients with conversion aphonia (CA) during symptoms and after recovery. CA is a rare conversion disorder characterized by sudden, enduring, nonorganic loss of voice, often

preceded by severe emotional distress. While resonant coughing and throat clearing is possible, indicating physically preserved vocal cord functioning, articulation in social contexts is virtually impossible. Despite controversy surrounding the precise etiological mechanisms, standardized circumlaryngeal manual therapy often results in rapid recovery after a single treatment [8]. This combination of a psychological etiology and a rapid nonpharmacological intervention make CA an intriguing model to explore associations between chronic stress exposure, dysregulations in the neural substrates of emotion processing and functional loss in neurological domains.

We used 2 fMRI paradigms to assess basal (emotional scenes) and higher-order (facial emotions) emotional processing in 2 female CA patients (19 and 36 years old) before and after successful intervention at the Phoniatics of the University Hospital Bonn. Two healthy female controls matched for age and education acted as controls and were administered the same therapy. Both patients suffered from enduring loss of voice (>4 weeks) following considerable emotional distress. Videolaryngoscopy evidenced normal contraction and relaxation of inner laryngeal muscles during respiration and coughing but pathological supraglottal constriction during phonation. Both patients reported substantial current stress accompanied by a history of adverse experiences due to parental dysfunction (physical abuse, emotional neglect) in one case and a worrisome and burdening relationship (divorce after 10 years, unfulfilled desire for children, miscarriage) in the other. Interestingly, both patients further reported to be generally "eager to please everybody" and to have severe problems "voicing their own feelings and needs". In both cases, the voice function fully recovered after treatment, thus enabling a direct pre-/posttreatment comparison of amygdala-prefrontal functioning on the same day, facilitating a thorough control of a range of potential confounders [for a similar approach, see 3].

Across both paradigms, the patients, but not the healthy controls, displayed increased activity in medial prefrontal regions as well as decreased emotional reactivity in the amygdala during symptoms relative to recovery (Fig. 1a). This pattern has previously been associated with emotion regulation success [9]. Interestingly, the lingual gyrus, a region with bidirectional pathways to the amygdala and associations to emotional reappraisal and suppression [10], also displayed altered functioning during symptoms, although the specific pattern of neural changes varied across tasks, showing higher activation upon scenic stimuli but lower activation upon neutral faces. Based on these findings it may be hypothesized that the CA patients might have initially gained control over their negative emotional state via top-down control, which successfully reduced negative emotions and associated amygdala hyperactivity. However, in the long run, such exaggerated top-down control may have disrupted the integrity of the amygdala-prefrontal circuit leading to loss of normal function, in this case, voice loss.

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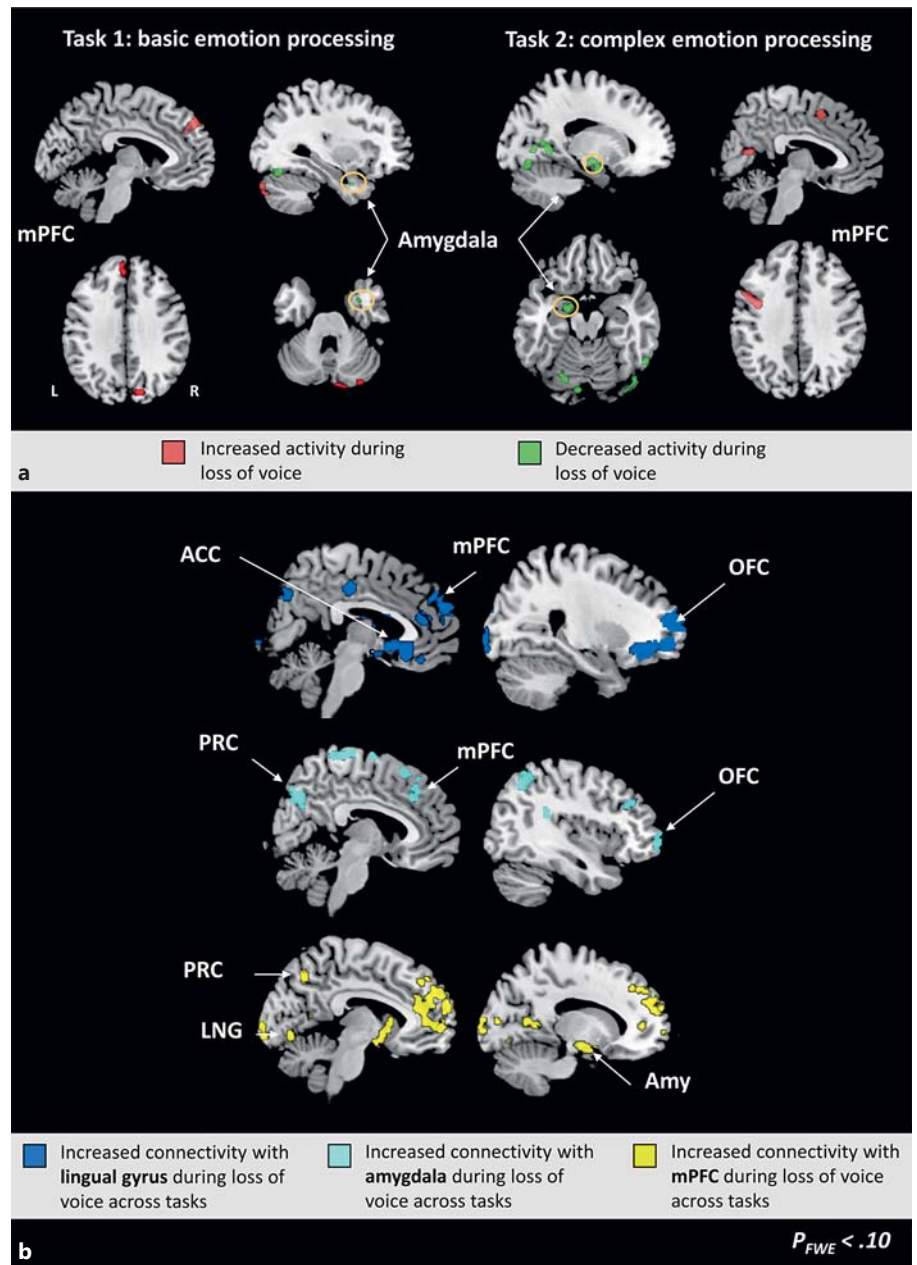


Fig. 1. Neural alterations in the emotion-processing networks during symptoms. Neural response (**a**) and functional connectivity (**b**) in patients during symptoms relative to recovery. Axial sections are presented in neurological orientation, left-side left. ACC, anterior cingulate cortex; Amy, amygdala; L, left hemisphere; LNG, lingual gyrus; mPFC, medial prefrontal cortex; OFC, orbitofrontal cortex; PRC, precuneus; P_{FWE} , family-wise error corrected p value; R, right hemisphere.

To specifically examine pathological inhibition patterns, i.e., exaggerated and inflexible emotion regulation underlying CA, we conducted a functional connectivity analysis. Results further supported the hypothesized mechanism by revealing increased bidirectional coupling of both amygdala and lingual gyrus with extensive prefrontal regulatory networks and further areas involved in emotional reactivity, including striatal regions (Fig. 1b) during voice loss as compared to recovery. Again, controls did not display comparable neural changes following treatment.

Taken together, the symptomatic state was characterized by increased involvement of medial prefrontal regions and concomitantly decreased amygdala emotional reactivity, while connectivity

between both regions was enhanced. Importantly, these pathological patterns in the amygdala-prefrontal circuit normalized after circumlaryngeal therapy and recovering of voice quality. Interestingly, stress-related disorders, including PTSD, have been associated with deficient emotion regulation and reduced governance of the prefrontal cortex over the amygdala in the context of hypoconnectivity in the amygdala-prefrontal circuitry [6, 7] whereas we observed exaggerated top-down control in the CA patients, suggesting that emotional distress might trigger divergent psychopathological disruptions in the amygdala-prefrontal circuit. Examining the contextual and risk factors that promote the specific symptomatology in response to exaggerated stress seems a fasci-

nating research question for future studies. Given the exploratory nature of the present case report, the hypothesized neuropathological mechanism underlying CA needs to be considered with caution, and findings need to be validated in larger samples.

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Disclosure Statement

The authors report no competing biomedical financial interests or personal affiliations in connection with the content of this manuscript.

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