

Case Report

“Gourmand syndrome” in a child with pharmacoresistant epilepsy

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ABSTRACT

We report the case of a 10-year-old boy with pharmacoresistant epilepsy, symptomatic of a right temporoparietal hemorrhagic lesion, who displayed an eating passion as described for the gourmand syndrome (GS) in adults and discuss the role of epilepsy in GS. This patient presented with a significant change in his eating habits (abnormal preoccupation with the preparation and eating of fine-quality food) concordant with the onset of his seizure disorder, without any previous history of eating disorders or psychiatric illness. This observation corroborates the important role of the right cerebral hemisphere in disturbed eating habits, including the relatively benign GS, and, possibly rarely, in less benign eating disorders such as anorexia and obesity.

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1. Introduction

Eating disorders rarely develop from brain lesions in hypothalamic, brainstem, and hemispheric sites [1]. If the lesion resides in the cerebral cortex, the right hemisphere is often incriminated. *Gourmand syndrome* (GS) has been described as a benign eating disorder associated with lesions most often involving the right frontal and temporal structures. It involves a preoccupation with food and a preference for fine eating, and was first reported in 1997 in adult patients subsequent to cerebral lesions [2]. GS related to a seizure disorder, however, has not yet been reported in young children. We report the case of a child with pharmacoresistant epilepsy, symptomatic of a right temporoparietal hemorrhagic lesion, who displayed an eating passion as described for the GS in adults. The role of epilepsy in GS is also discussed.

2. Case

The patient, a right-handed 10-year-old boy, was born at 34 weeks of gestation after an uneventful pregnancy. He suffered from streptococcus B sepsis and hemorrhage in the right temporal lobe during his neonatal period. However, his development was normal

with no particular events until the age of 8, when he started having seizures. The seizure semiology is stereotyped, with loss of contact and, rarely, oral automatisms or dystonic posturing of the left hand. He has never had any generalized tonic-clonic seizures or febrile convulsions. Seizure frequency varies from once a week to seven times per day, with little improvement despite treatment with different drugs. Pharmacoresistance was established against five different antiepileptic drugs (AEDs).

The parents noticed a change in his eating habits within the same year of seizure onset. His appetite increased, but his preference for fine food was more striking. He would cook for himself and for the family, especially on weekends, and often became preoccupied with development of new recipes. For example, he was very proud of his “Reispfanne” (rice preparation), a new recipe he created by himself that certainly tasted delicious according to his parents. They reported that he had preferences for certain sandwiches, noodles, fish, and lamb dishes and certain fruits (sour fruits, grapefruit, oranges, etc.), but did not like fried foods or chips. Although it was popular among his peers to go to fast food restaurants, he would insist that he did not like the quality of food there and preferred to cook for himself. Although his father runs the bakery in his home town, he was not exposed to any specialized or refined cooking within the family. In his family as well in the other families living in this part of Switzerland, traditional gender role assignment implied that cooking was done by women (as it was in the patient's family). He has a brother (5 years) and a sister (6 years) in good health without any particular interest in eating or

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cooking. There was no history of binge eating, anorexia, bulimia, purging behavior, or any preoccupation with his weight. He is an emotionally stable, friendly boy with good communication skills. He did not undergo a formal psychiatric examination; however there was no history of affective disorders, nor were any symptoms apparent during the period of hospitalization.

His family history is negative in terms of neurological or psychiatric disease or mental retardation. There is no history of eating disorders (anorexia, bulimia, etc) in any family member. Because of persistent seizures, he was referred to our unit for the presurgical evaluation. He was overweight with a BMI of 25, (more than 2 SD, > 97, for swiss children). Neurological examination revealed left inferior quadrantanopia and left dysidiadochokinesia. Previous EEGs showed a right posterior focus. During the evaluation period, a total of 19 seizures were recorded, the seizures were characterized by right parieto-occipital onset or, if occurring during sleep, by a more diffuse bilateral parasagittal onset. Magnetic resonance imaging examination revealed the sequelae of right temporal hemorrhage with porencephaly and periventricular gliosis as well as hemosiderin deposits in the right parietal lobe (Fig. 1). Neuropsychological examination suggested right hemispheric dysfunction (visuospatial memory deficits, discrete signs of neglect, figural fluency), more pronounced during the postictal phase (additional perseveration and confabulations).

3. Discussion

This 10-year-old boy presented with a significant change in his eating habits when his seizure disorder started at the age of 8, without any previous history of eating disorders or psychiatric illness. He developed a passion for eating good food, preparing certain types of food, and inventing new recipes, simultaneous with the onset of his epilepsy disorder. Thus, his eating behavior fits very well into the spectrum that has been described as gourmand syndrome strongly associated with right-sided brain lesions [2].

In our patient, although the frontal lobe appeared both anatomically and functionally intact (PET, ictal, interictal EEG), he had a

large temporal lesion. In the first large case series reported [2], most patients with GS had a frontal or temporal lesion. In fact, 30 of 36 also had temporal damage (83%), with significant predominance of the right side. Twelve patients had additional parietal damage (33%). In 34 patients (94%) the right hemisphere was involved. Thus, in this series of adult patients with GS, despite the predominance of right anterior lesions, right temporal and parietal lesions were found as well and are in line with the notion of a right hemispheric disorder in GS.

Interestingly, 10 patients (22%) also had epilepsy, as did our patient. In a recent review of eating disorders (anorexia, bulimia) related to cerebral lesions, 12 of the 13 cases (92%) had epilepsy [1]. It may be speculated that neuronal activity beyond the lesion proper (as in epilepsy) triggers the establishment or activation of a network that finally results in the development of complex eating behavior and attitudes toward eating. This may also explain why only 7% of patients with focal cerebral lesions develop GS [2]; that is, recruitment of remote structures may be needed in some cases to develop changes in eating habits. Concordant with this concept, our patient developed the GS when the epilepsy started, although the lesion had been present since birth. Moreover, the eating disorder has been shown to be reversible in some/most cases when the epilepsy is controlled [3–6].

There are two major aspects of GS: (1) the food and eating habits and aberrant food consumption behavior; (2) the obsessive component, which may result in craving and perseveration. The most common eating disorder today is obesity, and the proportion of obese people is constantly rising in Western societies. Similar to GS, anorexia, or bulimia, there is evidence suggesting that subtle neuronal dysfunctions might play a role in obese subjects who have an anatomically intact brain [7]. The right prefrontal cortex mediates decision making and perception of body self-image, which are all possibly crucial in the development of obesity. Discrete right prefrontal dysfunctions have been demonstrated in obese subjects by behavioral and fMRI studies [8].

Our patient is also obese. Although there may be additional right frontal dysfunction resulting from propagation of epileptic

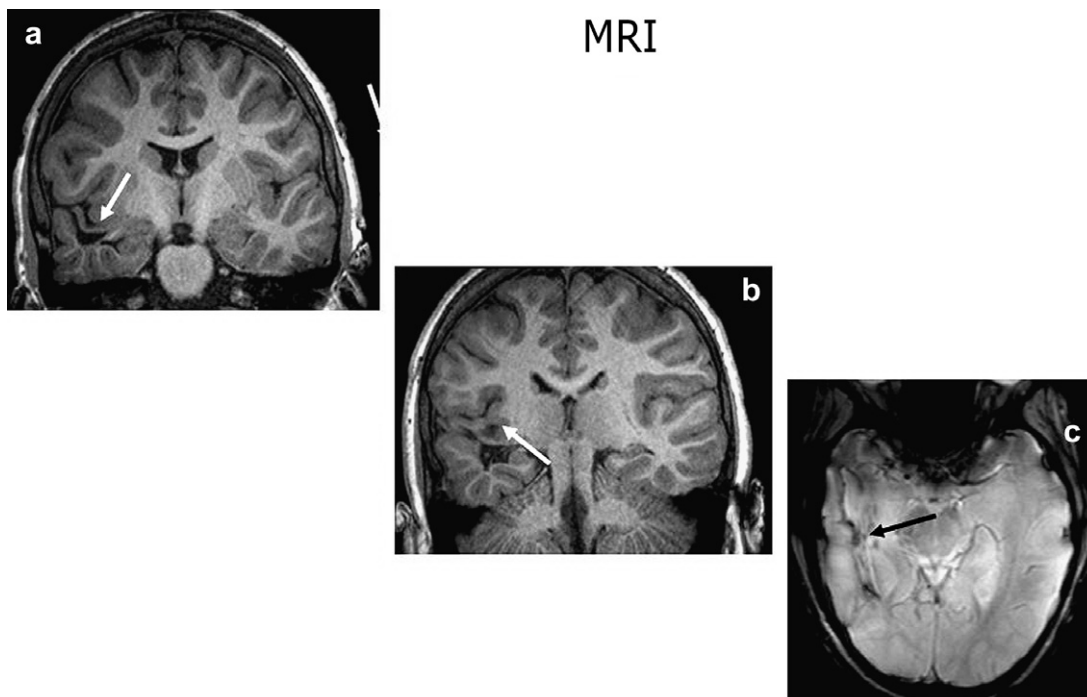


Fig. 1. Three-dimensional T1 coronal slices (a, b) show the sequelae in the right temporal gyrus (arrows) with hemosiderin deposits on gradient echo T2 (arrow c).

activity to these structures, as suggested by the ictal EEGs and the neuropsychological deficits, obesity may also be the result of a long-lasting GS. Obesity (BMI 28–30) was noted in 19% of adult patients with GS [2] compatible with the range noted in our patient, taking the corresponding age BMI values into account. Thus, obesity may represent a second eating disorder in our patient, or may be part of GS, both resulting from a right hemispheric disorder.

GS also relates to the obsessive and passionate aspects of eating. There is a large body of research on the molecular and anatomical aspects of craving, withdrawal, and binge behavior in the context of drug addiction [9]. Although the aforementioned symptoms are not part of GS, these patients often describe a preoccupation with food, that is, craving and expectation of the desired stimulus (“fine food”). This aspect of addictive behavior seems to be supported by the orbitofrontal, prefrontal, and cingulate cortex. Although no focus was noted in these structures in our patient, it could be speculated that they are easily recruited by the temporo-parietal epileptic activity.

Disturbed eating habits may not only be of psychosocial origin, for example, through family-based pathological eating habits, but also may be related to right hemispheric brain dysfunction. Alterations in eating habits and weight gain or loss are described as side effects of a variety of AEDs such as valproate, topiramate, and zonisamide; however, these drugs may not be causal in all patients. In our patient, GS (and obesity) did not change while he was on five different AEDs including topiramate. The onset of disturbed eating habits at an unusual age or in an unlikely gender, together with a history of head injury or epilepsy, should prompt neurological examination, including brain imaging. As many eating disorders start in childhood and adolescence and, with the exception of GS, are not benign, more research in this domain should elucidate underlying brain mechanisms and help to develop alternative treatment strategies.

Ethical approval

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

Conflict of interest statement

None of the authors has any conflict of interest to disclose.

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