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Constellation of psychic symptoms in one patient with mesial temporal lobe epilepsy with hippocampal sclerosis and the artistic representation of the patient's pre and post surgical experiences

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Abstract

Introduction: We describe a constellation of psychic symptoms in a professional painter with mesial temporal lobe epilepsy and hippocampal sclerosis, rarely described together in one patient, as well as the patient's artistic representation of these symptoms.

Case presentation: Prior to surgery, our patient experienced visual hallucinations, precognitions, out-of-body experiences, right unilateral mydriasis, and severe headaches, which led to a diagnosis of drug-resistant temporal lobe epilepsy. After surgery, she developed a visual field defect, retrograde trans-synaptic degeneration, anomic aphasia, foreign language syndrome, mirror writing, and Geschwind syndrome.

Discussion: Each symptom is presented in comparison with previous reports of patients with similar symptoms related to temporal lobe pathology.

Conclusion: Patients with temporal lobe disease may suffer from numerous and varied symptoms that often go unrecognized by physicians due to their unusual presentation. Young epileptic patients with temporal lobe disease may also express these symptoms through their artistic production.

Keywords: Anomia, Temporal lobectomy, Depersonalization, Epileptic syndromes, Hallucinations, Hemianopsia, Hippocampal sclerosis, Illusions, Personality, Retrograde degeneration.

Constelación de síntomas psíquicos en una paciente con epilepsia mesial del lóbulo temporal con esclerosis del hipocampo y la representación artística de las experiencias de la paciente pre y post cirugía

Resumen

Introducción: describimos una constelación de síntomas psíquicos en una paciente pintora profesional con epilepsia mesial del lóbulo temporal y esclerosis del hipocampo, raramente descritos en conjunto en un solo paciente, así como la representación artística de sus síntomas.

Presentación del caso: previo a la cirugía, la paciente presentó alucinaciones visuales, precognición, experiencias extracorpóreas, midriasis unilateral y cefaleas severas, las cuales finalmente llevaron al diagnóstico de epilepsia fármaco resistente del lóbulo temporal. Luego de la cirugía, la paciente desarrolló defectos del campo visual, degeneración retrógrada transináptica, afasia anómica, síndrome de lenguaje foráneo, escritura en espejo y síndrome de Geschwind.

Discusión: cada síntoma se presenta en comparación con reportes previos de pacientes con síntomas similares relacionados con enfermedad del lóbulo temporal.

Conclusión: los pacientes con enfermedad del lóbulo temporal pueden sufrir una gran variedad de síntomas que suelen pasar desapercibidos por los médicos debido a su inusual presentación. Los pacientes jóvenes epilépticos con enfermedad del lóbulo temporal pueden, además, expresar dichos síntomas a través de sus representaciones artísticas.

Palabras clave: anomia, lobectomía temporal, despersonalización, síndrome epiléptico, alucinaciones, hemianopsia, esclerosis hipocampal, ilusiones, personalidad, degeneración retrógrada.

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Introduction

A 41-year-old female patient from a low socio-economic status presented with terrifying hallucinations, out-of-body experiences, precognitions, right unilateral mydriasis, and severe headaches. At the age of four, without any type of relationship with art, she decided that she would become an artistic painter. Her illness began at 5 years of age, marked by a constellation of rare symptoms that continued for many years until her diagnosis was established. With the intention of contextualizing the artistic representations in paintings of the symptoms suffered by the patient, we present her detailed symptomatology as described by the artist.

Hallucinations: These were described as present in the "right eye" accompanied by severe left hemicranium headaches. She recalled four specific, terrifying episodes. The first happened at the age of five, when she saw a deceased girl in her father's arms at the door of a church. The girl did not move but suddenly opened her eyes, which were completely white. At that same moment, our patient started to see on her left shoulder images of angel heads without hands or feet, floating freely in the air, similar to the Victorian-era depictions of angels. Given the terrifying nature of the hallucinations and the images that she painted seeking to artistically express her condition (Figure 1), her family soon believed she was possessed by demons and forced her to consult three priests, who, after studying the case, concluded, "She is the kind that can see them, but they do not penetrate her."

Precognition: She recalls lucid episodes in which she dreamed of numbers or catastrophic images that days later would come true. On one occasion, she played the numbers in a local lottery and won; however, her catastrophic dreams did not have such happy endings. In one of them, she remembers a plane crash involving children, and the following day she would read in the newspaper that it had actually happened. She still keeps the newspaper clipping.

Out-of-body experiences: She remembers how her soul came out of her body, and she saw herself lying on her couch.

Headaches: During that time, she experienced multiple severe headaches that became increasingly frequent and intense, often accompanied by choppy images in her right visual field (Figure 2.A) and, on some occasions, by loss of consciousness. Some

Figure 1. Pre-surgical drawings and paintings made many years before diagnosis

Note. A) Strange figures, described as "demoniac images", coming out from the head, showing what could be hallucinations at an early age. **B)** Interestingly, in many of her pictures or drawings, the left hemicranium is painted with dry branches coming out from the head on the same side of the epileptic focus, years before she was diagnosed. In this particular drawing the contralateral hand is burning. **C)** This is the representation of a normal person with a headache (broken head), visual symptoms (the eye is pierced by a snake-like figure), and different faces coming out of the head that could resemble out-of-body experiences. **D)** Eyes everywhere, some of them cracked, others crying in a blue dimension. Interestingly, the patient told us that before surgery, most of her paintings were predominantly blue because she was fixated on that color, reminiscent of another famous post-impressionist epileptic painter, Vincent van Gogh, who was fascinated with cobalt blue.

Source: With permission from the artist-author Angela Rojo.

episodes were very disabling and refractory to painkillers. She was struck by the fact that her right pupil dilated for hours and improved spontaneously at dawn, a phenomenon that she also demonstrates in some of her paintings (Figure 2).

She underwent several EEGs from 2005 to 2014, which were initially inconclusive in relation to the epileptogenic focus. The last scalp EEG finally demonstrated focal paroxysmal abnormalities in her left temporal lobe. Due to complex side effects and poor response to different treatments, she was diagnosed with drug-resistant temporal lobe epilepsy. Her headaches became so intense that she once asked one of her specialists if they could remove that part of her brain. To plan surgery, it was imperative to locate the epileptic focus.

Presurgical evaluation included brain MRI, which showed no structural damage. Fluorodeoxyglucose (FDG) positron emission tomography (PET) (FDG-PET scan) revealed hypometabolism in the most lateral aspect of the medial gyrus of the left temporal lobe. A continuous 120-hour video-encephalographic recording (CV-EEG) demonstrated abundant interictal irritative activity in the middle-posterior left temporal lobe (T3-T5), with ictal progression to the left temporal-posterior occipital region. These findings supported the diagnosis of focal epileptic syndrome and ultimately completed the paraclinical workup before surgery.

Prior to surgery, the patient does not recall being hospitalized for the invasive monitoring electrode placement, nor do her medical records mention an invasive EEG procedure. However, the patient does recall being informed of the potential risk of permanent memory, speech, and visual field deficits. Therefore, she painted a series of drawings to express her fears (Figure 3). Interestingly, some of these drawings were done years before she even knew she had a disease.

In March 2017, she underwent a left temporal lobectomy extended to the occipital region and an amygdalo-hippocampectomy. She remembers that in the early postoperative period, while she was alone in her room, she saw a person sitting on her bed who was looking at her without speaking. Months later, when visiting a church, she would recognize the face of the person who appeared in that dream as Saint Francis of Assisi. This would be the last of her visions.

Figure 2. Pre-surgical drawings and paintings depicting the patient's headache patterns

Note. A) This is probably one of the most representative images prior to surgery, in which she clearly expresses headaches. She painted horns on the same side of the epileptic focus, without knowing it, and a horn cut into pieces coming out from the eye, illustrating the fragmented images she used to see in her right visual field. **B)** She was struck by the fact that her right pupil dilated (anisocoria) for hours and improved spontaneously at dawn. Even after surgery, this phenomenon still occurs occasionally, but because it resolves spontaneously, it is difficult to capture in a regular medical appointment. We asked her to send us a photograph when this happened, which she did by using her phone against a mirror; this is why in panel B the mydriasis appears in the left eye. **C)** and **D)** both represent this mydriasis, consistently shown in the right pupil.

Source: With permission from the artist-author Angela Rojo.

After the surgery, she never experienced similar images again. She reports being completely cured of her hallucinations, out-of-body experiences, sense of presence, and headaches.

She has retained the ability to paint despite having only a left visual field (Figure 4), as surgery resulted in a right homonymous, incongruent, incomplete hemianopsia. In our consultation, we observed a bow-tie pattern of optic nerve fiber atrophy in the right eye and atrophy of the nerve poles in the left eye, findings that were confirmed with optical coherence tomography (Figure 4). Her post-surgery paintings

Figure 3. Drawings created before surgery illustrating the patient's fears of collateral damage

Note. Prior to surgery, her doctor warned her that she would likely have problems with memory, speech, and parts of her visual field. In response, she created a series of drawings alluding to this situation as a way to express her fears. **A)** The eyes are covered by a band, and the face has no mouth. **B)** Here the mouth is covered by a band. **C)** There is a nail in the right forehead (the surgical procedure from Angela's point of view), and the mouth is sutured, reflecting her feelings and fears about surgery. **D)** This is kind of an angel-like figure without a mouth but with just one left wing; the absence of the right wing symbolizes her sense of being broken in two (a theme also expressed in many of her other drawings and paintings). Interestingly, some of these drawings were done years before she was diagnosed with epilepsy.

Source: With permission from the artist-author Angela Rojo.

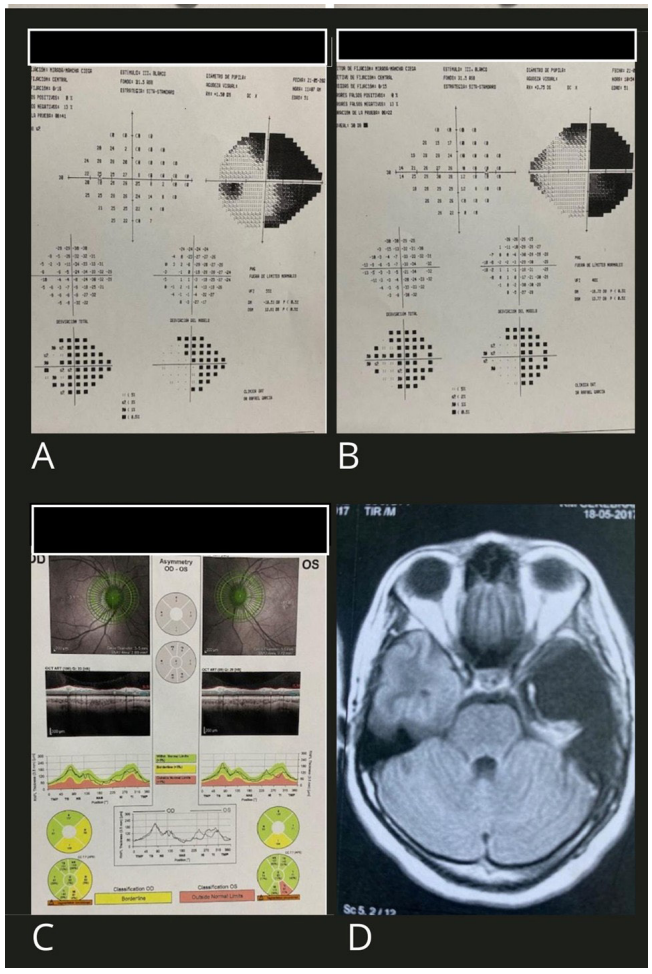

Figure 4. Ancillary studies demonstrating functional and organic damage after surgery

Note. A) Left and B) almost congruent right hemianoptic defect after left temporal lobectomy. **C)** Optic nerve coherence tomography showing borderline thinning of the retinal nerve fibers at the inferior poles of the left optic nerve (inferior direct fibers) and a borderline bow-tie pattern in the contralateral right optic nerve due to thinning of the nasal fibers (cross fibers) due to retrograde transsynaptic axonal degeneration. **D)** T1 brain axial MRI showing no left temporal brain after surgical lobectomy.

Source: With permission from the patient.

depict dry, dead branches emerging from the eye on the side of her visual field loss, while on the contralateral side she portrays a skull with wounds (Figure 5). In several of her more recent paintings, she paints branches growing again from her head, symbolizing neuronal resurgence (Figure 5D).

Since surgery, she has had to learn to rename words (anomic aphasia), and to achieve this, she often resorts to circumlocution. For example, when she was not able to say the word "compact disc" or "CD", she made herself understood by saying, "the round, flat, thing that you put inside the computer and has information".

Figure 5. Artistic representations of reality and hope following surgery

Note. In many of her paintings after surgery, she attempts to depict signs of renewal and neuronal resurgence. **A)** Before surgery, she painted an angel with a broken left head and just one wing. **B)** After surgery, she painted a dry tree branch emerging from the left side (left lobectomy) but included a white feather-shaped tear on the right side as a symbol of resurgence. **C)** Before surgery, she painted an angel-like figure with scaly skin only on the left side, a single wing, and a dry tree on the right side of the head. **D)** After surgery, she painted trees in bloom that had previously appeared dry. A similar pattern can also be observed in Figure 1C.

Source: With permission from the artist-author Angela Rojo.

It was surprising for us that during her first consultation she was able to say some phrases in Latin and English (foreign language syndrome), and when asked if she already spoke those languages, she responded "well, I imagine that maybe I must have learned something from them at some point, but I really do not remember." Although she finds it difficult to read —and after a relatively short reading, she feels as tired as when she had the post-ictal periods— she finds it more comfortable to read and write from right to left (mirror reading and mirror writing, respectively). She is able to do this with both hands but does not perceive her visual environment, faces, or drawings as inverted. Regardless of the direction she reads or the hand with which she writes, she does not remember what she has read or written.

Further investigation was conducted, including a home visit to review her artwork. During the visit, she told us that three different men had proposed to her, but she felt like "a disgusting woman" because of it. She also showed us notebooks filled with a distinctive handwriting style (Figure 6). Many of the drawings were related to angels and religious themes. It was concluded that, in addition to the strange variety of symptoms, she also met criteria for a diagnosis of Geschwind syndrome.

Discussion

Prior to surgery, the patient had a constellation of symptoms, including visual hallucinations, precognitions, out-of-body experiences, right unilateral mydriasis, and severe headaches, which finally led to the diagnosis of refractory temporal lobe epilepsy. After surgery, she suffered from visual field defect, retrograde trans-synaptic degeneration, anomic aphasia, foreign language syndrome, mirror writing, and Geschwind syndrome, a constellation of symptoms rarely described together in a single patient. Accordingly, we discuss each symptom.

Visual complex hallucinations, together with the interesting constellation of symptoms observed in our patient, fit within what was previously known as "psychic seizures", usually related to temporal lobe epilepsy (TLE) (1). Psychic seizures were described

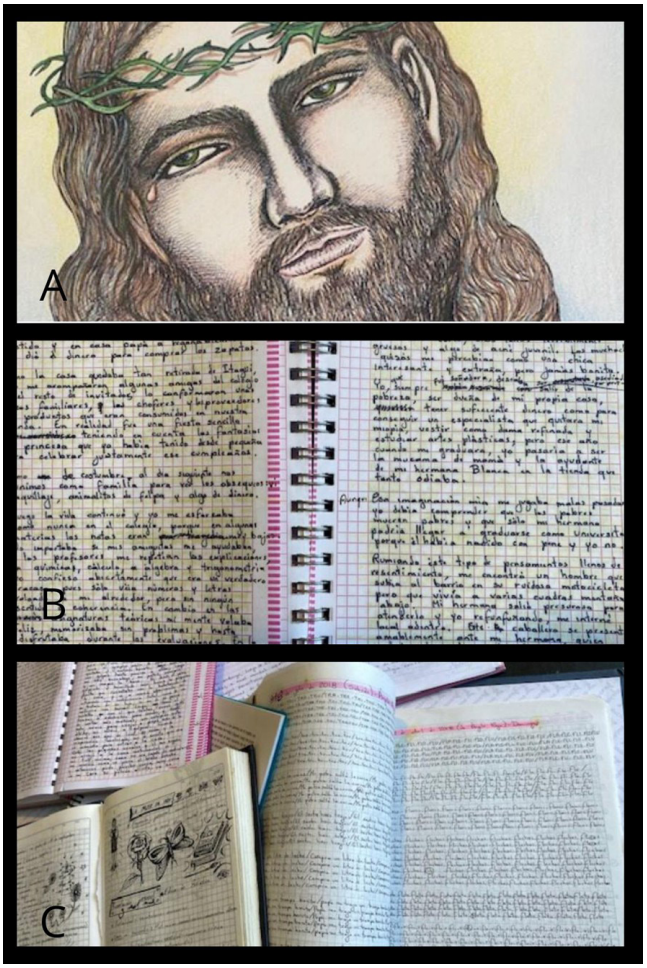

Figure 6. Features of Geschwind syndrome

Note. A) Recurrent religious paintings. B) Prior to surgery, the writing was dense, filling all of the available space on each sheet of paper. C) Examples of Geschwind syndrome's hypergraphia. The patient produced numerous pages of material, predominantly focused on religious themes and church-related topics. She obsessively preserved and archived her letters, filling entire drawers.

Source: With permission from the artist-author Angela Rojo.

as simple partial seizures presenting as psychological or mental phenomena. However, the terms "partial" and "psychic" have been replaced by "focal" and "cognitive", respectively. Most of these seizures last less than three minutes and are often associated with visual or auditory illusions and hallucinations, accompanied by emotional changes, such as agitation, fear, or paranoia.

Additional psychic experiences may include depersonalization, derealization, autoscopy phenomena, out-of-body experience, or the sensation of "someone behind" (2). Autonomic manifestations are also common. Unilateral pupillary dilation can occur and, in some cases, may help lateralize the seizure to the ipsilateral hemisphere (3,4). However, in our patient, the dilated pupil did not lateralize the origin of her seizures.

Surgical resection provides a 60% to 80% chance of seizure freedom at two years for patients with drug-resistant mesial temporal lobe epilepsy (MTLE), although long-term outcomes tend to be less favorable (5,6). Seven years after surgery, our patient has remained free of "psychic hallucinations" and disabling headaches. Anterior temporal lobectomy is generally considered safe, with the most common neurologic complications being visual field deficits and memory dysfunction, both of which are currently present in our patient. In her case, the surgeons performed a left temporal lobectomy with extension to the occipital region and an amygdalohippocampectomy. Due to the extent of the resection, the patient suffers from other unexpected complications.

Histopathological analysis revealed focal cortical dysplasia in temporal lobe sections and reactive gliosis with neuronal dyslamination in the hippocampus fragments, which led to the diagnosis of hippocampal sclerosis. According to the current epilepsy and seizure classification established by the International League Against Epilepsy (ILAE), she falls into the category of mesial temporal lobe epilepsy with hippocampal sclerosis (MTLE-HS) (3,4).

Visual field defects

Visual field defects (VFD), most often homonymous superior quadrantanopsias, are a common consequence of open anterior temporal lobectomy (ATL), with incidence rates ranging from 9% to 100% (7-10). The severity of these defects is positively correlated with the volume of tissue resected (11,12). Consequently, ATL leads to tissue damage that includes "collateral damage" to the optic radiations within the temporal lobe. Schmeiser et al. (9) reported a 9% occurrence of visual field defects following limited subtemporal resections, compared to 35% after standard ATL. In our case, due to the extension of the left temporal lobectomy, considerable damage to the contralateral right visual field is expected.

Trans-synaptic degeneration

It is widely recognized that any condition impacting the visual pathway anterior to the lateral geniculate body can lead to secondary optic atrophy as a result of retrograde trans-synaptic degeneration. Optical coherence tomography (OCT) scans of our patient revealed borderline thinning of the retinal nerve fiber layer at the inferior poles of the left optic nerve (inferior direct fibers) and a borderline bow-tie pattern in the contralateral right optic nerve due to thinning of the nasal fibers (crossed fibers) (Figure 4C). This optic nerve fiber pattern suggests that trans-synaptic degeneration is not limited to injuries involving the visual pathway anterior to the lateral geniculate body (13-17). Vries-Knoppert et al. (16) identified three distinct patterns of retrograde axonal degeneration using OCT in a study of twenty-five patients undergoing elective partial temporal lobe resection for intractable epilepsy. Retrograde trans-synaptic degeneration has also been demonstrated in patients undergoing occipital lobectomy for tumor excision (17).

Anomic aphasia

There are different types of anomic aphasia, and in clinical practice, a mixture of several varieties of anomia is frequent. The type relevant to our case is "disconnection anomia", which occurs due to the disruption of connections between the sensory and language cortices, specifically involving the white matter tract known as the arcuate fasciculus (18).

Patients with disconnection anomia may show modality-specific anomia, where the inability to name objects is restricted to a particular sensory modality, such as vision. For instance, a patient who can name an object when presented through other sensory modalities like hearing or touch may struggle to name the same object when seen. In this case, the anomia results from a disconnection between the visual cortex and language areas (19). If the patient is unable to find the appropriate word, they may resort to a different description, leading to a circular speech pattern known as circumlocution.

Interestingly, after surgery our patient was able to re-learn certain words, which means that there must be neuroplasticity mechanisms or alternative pathways that can be developed, trained, or activated after damage. Nardo et al. (20) discovered that both immediate and long-term improvements in naming were dependent on a shared bilateral neural network. This network included the anterior insula, inferior frontal cortex, and dorsal anterior cingulate cortex in the right hemisphere, and the premotor cortex and supplementary motor area in the perilesional left hemisphere. The activation of multiple bilateral regions may explain why our patient, and others with anomic aphasia, were able to successfully re-name words despite damage to the left temporal lobe.

Foreign language syndrome

During the interview, the patient surprised us by suddenly speaking phrases in both Latin and English. Sometimes, when a person with anomic aphasia is multilingual, they may unintentionally switch languages in an attempt to find the correct word they are missing. This is referred to as "inadvertent code-switching". However, our patient's case is particularly interesting, since she did not recall speaking these languages.

Is this a consequence of the cognitive (memory) damage expected from the surgery? This could be the case if she studied those languages in the past. While English is a common language, Latin is not, and given the patient's upbringing in a very poor environment, it is highly unlikely that she ever had the opportunity to learn it.

An alternative explanation is "foreign language syndrome", in which patients shift from their native language to focus on a second language for a period of time. This language switch usually happens after surgery and typically resolves on its own within a short duration, but in our patient these phrases were spoken seven years after surgery. The precise pathophysiology of foreign language syndrome is still unknown, particularly regarding whether it is a distinct syndrome or a phenotype of emerging delirium (21). There is still much to understand, and further research is required.

Mirror writing

Mirror writing is an uncommon form of script, where the text runs in the opposite direction of standard writing, with individual letters reversed, making it easier to read with the aid of a mirror (22-25). Certain parts of the brain have been (at least anatomically) related to mirror image equivalence (22-26). Four white matter areas in the brain interconnect the

right cerebrum hemisphere with the left cerebrum, but in mirror-image equivalence, the temporal lobe (infero-temporal cortex), the anterior commissure, and the hippocampus have been most specially involved. In our case, these structures were somehow affected due to the extension of the left temporal lobectomy, explaining at least anatomically the occurrence of mirror writing and reading. Among the various theories proposed to explain this unusual phenomenon, we consider the visual word-form hypothesis —initially introduced by Orton in 1928— to be the most relevant to our case. This theory suggests that bilateral, mirror-reversed visual engrams are formed in opposite hemispheres due to homotopically organized commissural connections. During reading development, mirrored graphemes in the non-dominant hemisphere (typically the right) are suppressed. However, under pathological conditions this suppression may be lifted, leading to the emergence of mirror writing and reading (26).

Geschwind syndrome (The interictal behavior syndrome)

Norman Geschwind proposed that limbic damage occurring during seizures is responsible for this syndrome (27,28). Others propose that chronic amygdala stimulation from seizure activity or bilateral temporal lobe dysfunction could also explain the interictal behavioral changes described by Geschwind (29-30). In our patient, characteristic symptoms such as hypergraphy, religiosity, hyposexuality, and a rigid personality were evident since the beginning of the disease and persist to this day. Following surgery, although she is no longer able to write as she used to, she continues to paint lots of angels. In fact, she once expressed, "I do not know where it comes, this fervent desire to paint angels".

Conclusion

Patients with temporal lobe epilepsy may experience more than one of these unusual symptoms. Recognition of such manifestations is often challenging for several reasons: sparse literature, unusual descriptions, strange psychological and religious relationships, patient's fears of being judged, and the fact that some features, previously included in the term "psychic seizure", have been abandoned (or at least not explicitly described) in the new ILAE classification.

Art has been utilized as a means to express what cannot be conveyed through words, and in our patient with anomic aphasia, this was the case. We consider the small sample of artwork shown here to be extraordinary, as it is probably the first time an epileptic artist can express through her paintings all her feelings and experiences across different stages of the disease, in a similar way to the famous Mexican artist Frida Kahlo.

Also, the artwork shown in this case has a potential scientific purpose. During the Latin American Summer School on Epilepsy (LASSE XIII) in 2020 (31), several participants' paintings displayed common patterns. Therefore, the brief collection of artwork shown in this case could help physicians recognize some pattern in paintings of epileptic patients, thereby fostering empathy and potentially facilitating earlier recognition in children who may be unable or afraid to verbally express their experiences.

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