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COMPLETE REMISSION OF EPILEPTIC PSYCHOSIS AFTER TEMPORAL LOBECTOMY

Case report

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ABSTRACT - We report a case of a female patient with refractory complex partial seizures since 15 years of age, recurrent postictal psychotic episodes since 35 which evolved to a chronic refractory interictal psychosis and MRI with right mesial temporal sclerosis (MTS). After a comprehensive investigation (video-EEG intensive monitoring, interictal and ictal SPECT, and a neuropsychological evaluation including WADA test) she was submitted to a right temporal lobectomy. Since then, she has been seizure-free with remission of psychosis, although with some persistence of personality traits (hiperreligiosity, viscosity) which had been present before surgery. This case supports the idea that temporal lobectomy can be a safe and effective therapeutic measure for patients with MTS, refractory epilepsy and recurrent postictal epileptic psychosis or interictal epileptic psychosis with postictal exacerbation.

KEY WORDS: epilepsy, mental disorders, psychosis, temporal lobe.

Remissão completa de psicose epiléptica de paciente submetida a lobectomia temporal: relato de caso

RESUMO - Relatamos o caso de uma paciente que apresentava crises parciais complexas desde a idade de 15 anos, episódios psicóticos recorrentes desde os 35, evoluindo para psicose crônica interictal refratária, em quem investigação pela ressonância magnética (RM) revelou a presença de esclerose temporal mesial (ETM) direita. Após investigação abrangente (monitorização intensiva pelo vídeo-EEG, SPECT interictal e ictal e avaliação neuropsicológica incluindo teste de WADA), a paciente foi submetida a lobectomia temporal direita. Desde então, ela está sem crises e a sua psicose remitiu, persistindo apenas alguns traços de personalidade (religiosidade, viscosidade) que ela já apresentava anteriormente. Este caso dá suporte à idéia de que a lobectomia temporal pode ser uma medida terapêutica segura e eficaz para pacientes com ETM, epilepsia refratária e psicose epiléptica pós-ictal recorrente ou psicose epiléptica interictal com agravamento pós-ictal.

PALAVRAS-CHAVE: epilepsia, transtornos mentais, psicose, lobo temporal.

Several mental disorders may occur in patients with epilepsy. Psychoses associated with epilepsy are among those of major concern to neurologists and psychiatrists. These psychoses are classified into periictal (of which most significant are the postictal psychoses) and interictal. Postictal psychoses have onset in the late postictal period (hours or days after) of an isolated seizure or of a cluster of complex partial seizures (CPS) with or without secondary generalization. Its presentation is typically pleomorphic, with delusions and hallucinations, loose associations, mannerisms and mood swings[1,2]. Prevalence is of 4% in cases of apparently non-refractory epilepsy[3] and of 7-18% in cases of refractory epilepsy[2,4,5]. Interictal psychoses are chronic and/or recurrent, with delusions, hallucinations, thought disorders, disorganized behavior, relative absence of catatonic symptoms, mood swings, apragmatism and emotional blunting (less intense than that observed in classic schizophrenia), may progress with persistence of psychotic symptoms, psycho-organic sequelae, suicide attempts, multiple hospitalizations and functional descent[6,7].

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Prevalence in general population studies varies from 2-7,1%[8-10], in general clinics from 0,8-9,2%[13-20] and in neurological or epilepsy clinics from 8,8-27%[21-26].

The purpose of this paper is to report the case of a patient with refractory epilepsy and epileptic psychosis and the value of therapeutic surgery such as temporal lobectomy in her treatment.

CASE

A 47 years old female with onset of epileptic seizures at 12 years of age and progressive worsening so that on occasion of her referral to PROJEPSI (Epilepsy and Psychiatry Project) in 1996 (when she was 42 years old) she had clusters of seizures (mainly during premenstrual period or under emotional stress) with up to 10 seizures a day, followed by several days with no seizures. She had been followed up in several medical institutions and had made use of many antiepileptic drug (AED) regimes without ever achieving satisfactory control, besides use of antipsychotic drugs for her associated mental disorder (a psychosis). Seizures followed a stereotyped sequence: "death distress", disturbance of consciousness, falls, dystonic posture of left arm, head version to the right, staring, verbal and oral automatisms and postictal mental confusion. Rarely there was secondary generalization with tonic-clonic convulsions. Usual duration of seizures was 2 – 5 minutes. The patient's mother told that after the onset of epilepsy, she became "fanatic about religion". Around 15 years of age she was baptized into the Adventist Church (all members of her family are non-practicing Roman Catholics), began going to Church daily and dedicated a major part of her day to prayer (she would kneel and cover her head in a veil for this) often causing problems with family members. Relatives describe her as critical or self-demanding and overconcerned with moral issues. The patient told she was uninterested in going out, marriage or sexual relationships. She planned to dedicate her life to her Church but was "constantly harassed by men interested in dating her." Around 35 years of age she suffered profound behavioral changes following clusters of seizures. She became restless, wishing to "run away from home" (the reason she reported for leaving her family was their lack of faith in God), and also seeing bugs and faces on printed fabrics and hearing "the humming of crickets in her ears" interpreted as "either something in my head or a temptation from the Devil". She became excited when speaking about religious subjects, went out naked into the street saying that she would "enter into the Kingdom of God naked and barefooted". She felt to be responsible for a special mission (though denying to be a saint or an important religious icon) and told that the "Song of the Lord had come out of her throat like a trumpet echoing through the four corners of the Earth, tearing the heavens and the depth of the seas", and that as she opened the Bible with the "hands of God" and read a passage that said "Blessed are they who knoweth the sound of trumpets!" she realized that "God spoke through her". This religious ecstasy led her to change from the Adventist to the Evangelical Church. Occasionally she woke up at night crying out and praying, waking her family members and accusing them of sinfulness. After a few days, these manifestations would improve and then recrudesce after a new cluster of seizures. After some episodes she developed an interictal chronic psychosis with persistence of delusional beliefs, with worsening after clusters of seizures. She also had periods of depressive mood, anhedonia, inappetence, unstable affect, and frequently a feeling of lack of feelings. On these occasions she said she felt nothing, as if she were "a broken vase" and that she was no longer the same person, not feeling the same faith as before or even love for her own mother. Although she reported maintenance of lack of feelings, she wept a lot saying she was already dead and that she had felt happier and more alive when she had had seizures.

Phenitoin (up to 400 mg/day), phenobarbital (up to 200 mg/day), carbamazipine (up to 1600mg/day, leading to excessive sedation), valproic acid (up to 2500 mg/day) and clobazan (up to 30 mg/day) were used, in mono or polytherapy, with inadequate response. She maintained around 4 complex partial seizures per month, with sporadic secondary generalization. New generation AED were not available in the service. There were some problems with adherence to medication i.e. the patient would reduce dosage on her own when there was worsening of psychotic symptoms, either because of somnolence or because the AED were "the Devil's medicine". Haloperidol up to 17,5 mg/day, trifluoperazine up to 5 mg/day and risperidone up to 2 mg/day were sequentially used but she maintained her delusional beliefs. The patient had intense extrapyramidal side-effects (parkinsonian tremor) with only partial attenuation with use of biperidene up to 6 mg/day. Sertraline up to 100 mg/day was associated and some improvement was observed in depressive symptoms.

The following diagnoses were made according to the ICD X: symptomatic epilepsy with complex partial seizures (G.40.2), organic hallucinosis (F.06.0), organic delusional disorder (F.06.2) and organic personality disorder (limbic system epilepsy psychosyndrome (F.07.0).

As her improvement was not permanent and the course of psychiatric problems partially dependent on the epilepsy, which had itself inadequate control, from January 1999, when the patient was around 45 years old, the possibility of a surgical approach (right temporal lobectomy) was discussed. Despite the lack of insight into the unreality of her delusions, she demonstrade insight into the intractable nature of her seizure disorder. Both the patient and her mother gave informed consent for surgery.

Structural imaging and functional investigations were carried out such as MRI (Fig 1), interictal and ictal single photon emission computerized tomography (SPECT), routine electroencefalograms (EEGs), intensive video-EEG monitoring, neuropsychological evaluations and Wada

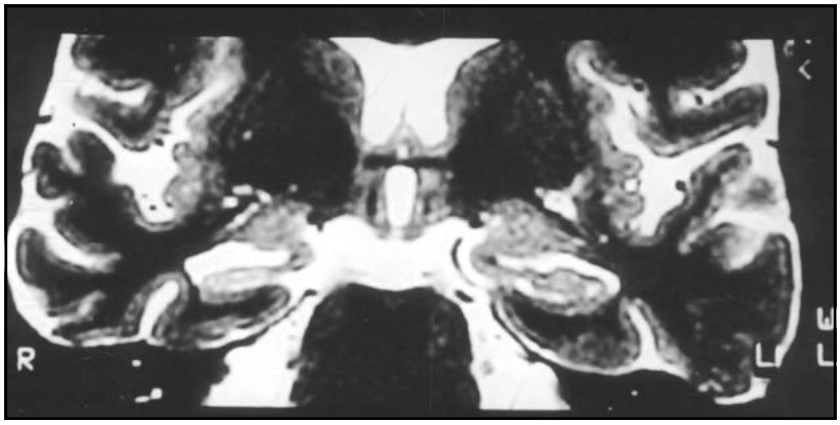

Fig 1. Pre-surgery MRI from the patient: coronal T2.

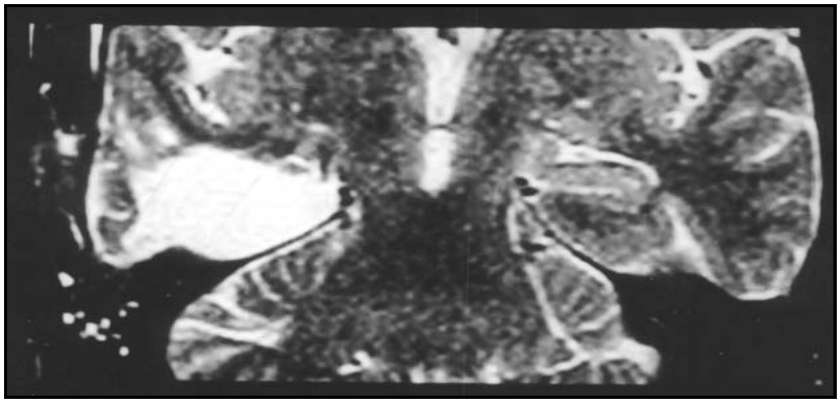

Fig 2. Post-surgery MRI from the patient: coronal T2.

test. These ancillary exams disclosed an epileptic zone in right temporal lobe and right mesial temporal sclerosis.

The patient underwent a standard right temporal lobectomy in August 11 1999 and was discharged from the neurosurgical ward in August 17 and has had an excellent postoperative outcome (Fig 2). She had only 3 epileptic seizures in the first weeks after surgery, progressing seizure free thereafter. She is still receiving treatment with carbamazepine 800 mg/day, clobazan 30 mg/day, risperidone 3 mg/day and biperidene 2 mg/day. Currently, one year after surgery, her personality traces are attenuated and she does not become as excited as before when discussing religious subjects, though she is still very devout. She has been euthymic with complete remission of psychosis for over one year.

DISCUSSION

Our case demonstrates that although postictal psychoses are of episodic nature, they may be recurrent with possibility of interictal transformation as observed by other authors[1,2,7]. In other words, psychiatric symptoms which at the beginning of the problem have a close temporal relationship with epileptic seizures become chronic and independent of them.

The association of psychoses with temporal lobe epilepsy (TLE) is reinforced by the presence of frequent CPS[6,7,27], seizures of limbic origin[28,29] and comorbidity with personality changes associated to TLE[6,9], all of them present in the case we have described. Guerrant et al apud McKenna et al.[30] and Small et al.[31] observed higher incidence of psychosis among patients with TLE but their samples were not large enough for statistically significant results. Shukla et al.[32] studied two comparable groups of 62 patients with TLE and 90 patients with generalized epilepsy and found significantly higher incidence of psychosis in patients with TLE. Gibbs and Gibbs[14] in a large study with 11000 patients studied 1675 patients with TLE and found a prevalence of psychosis of 12% as compared with a lower prevalence of 1% in 6671 patients with generalized epilepsy. Although not entirely conclusive, available evidence points to an association between TLE and psychoses.

Other aspects extensively studied are the nature and lateralization of cerebral disorders. Lesions of embryologic origin such as hamartomas and cortical dysgenesis[23,24] or hippocampal sclerosis, also known as mesial temporal sclerosis (MTS)[7,34] have

been suggested as risk factors for epileptic psychosis. Although there is evidence that pathologic processes situated in the left cerebral hemisphere as shown by left epileptic focus[27,35,36], left-handedness[23,26] or left MTS[7] are associated with epileptic psychoses, this is not invariably observed. There are reports of postictal psychoses accompanied by elation and hiperreliogisity associated with right hemisphere epileptic focus[37,38].

Several surgery programmes reject patients with chronic psychosis[39]. This decision appears to be based on the lack of effect of surgery on psychosis in most patients[6,24,40,41], on rare reports of postoperative psychiatric deterioration[22,40,42-44] or on the fear that psychosis would prevent adequate preoperative evaluation and postoperative rehabilitation. More recently, Fenwick has advocated surgical treatment in these cases on the basis that freedom from seizures is worthwhile despite unrelenting psychosis[45]. There is a number of studies that suggests a good outcome for postictal psychotic episodes after temporal lobectomy[45-48].

Although we recognize the limitations of this study (one case report only), the postoperative outcome of our patient suggests that temporal lobectomy may be a safe and effective therapeutic measure in patients with MTS and interictal psychosis which present postictal exacerbation

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