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Dacrystic seizures: Demographic, semiological and etiological insights from a multicenter study in long-term Video-EEG monitoring units

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Abstract

Purpose: To provide an estimate of the frequency of dacrystic seizures in Video-electroencephalography (EEG) long-term monitoring units of tertiary referral epilepsy centers and to describe the clinical presentation of dacrystic seizures in relationship to the underlying etiology.

Methods: We screened clinical records and video-EEG reports for the diagnosis of dacrystic seizures of all patients admitted for video-EEG long-term monitoring at five epilepsy referral centers in the United States of America and in Germany. Patients with a potential diagnosis of dacrystic seizures were identified and their clinical charts and video-EEG recordings were reviewed. We included only patients with: (1) stereotyped lacrimation, sobbing, grimacing, yelling

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or sad facial expression, (2) long-term video-EEG recordings (at least 12 hours), and (3) at least one brain MRI.

Key findings: Nine patients (four females) with dacrystic seizures were identified. Dacrystic seizures were identified in 0.06 to 0.53% of the patients admitted for long-term video-EEG monitoring depending on the specific center. Considering our study population as a whole the frequency was 0.13%. The presence of dacrystic seizures without other accompanying clinical features was found in only one patient. Gelastic seizures accompanied dacrystic seizures in five cases and a hypothalamic hamartoma was found in all of these five patients. The underlying etiology in the four patients with dacrystic seizures without gelastic seizures was left mesial temporal sclerosis (three patients) and a frontal glioblastoma (one patient). All patients had a difficult to control epilepsy as demonstrated by the following facts: (1) at least three different antiepileptic drugs were tried in each patient, (2) epilepsy was well controlled with antiepileptic drugs in only two patients, (3) six patients were considered for epilepsy surgery and three of them underwent a surgical/radiosurgical or radioablative procedure. Regarding outcome, antiepileptic drugs alone achieved seizure freedom in two patients and did not change seizure frequency in another patient. Radiosurgery led to a moderately good seizure control in one patient and did not improve seizure control in another patient. Three patients were or are being considered for epilepsy surgery on last follow-up. One patient remains seizure free three years after epilepsy surgery.

Significance: Dacrystic seizures are a rare but clinically relevant finding during Video-EEG monitoring. Our data shows that when the patient has dacrystic and gelastic seizures the cause is a hypothalamic hamartoma. In contrast, when dacrystic seizures are not accompanied by gelastic seizures the underlying lesion is most commonly located in the temporal cortex.

Keywords

Hypothalamic hamartomas; Ictal crying; Magnetic resonance imaging; Seizures; Semiology

INTRODUCTION

Dacrystic seizures present with paroxysmal stereotyped crying (Luciano, et al. 1993, Tatum&Loddenkemper 2010). Semiological features include stereotyped lacrimation, grimacing, sobbing, sad facial expression, yelling, and a subjective feeling of sadness. The clinical expression ranges from isolated changes in facial expression without lacrimation to dramatic bursts of lacrimation, sobbing, and marked changes in facial expression (Davison&Kelman 1939, Efron 1961, Gascon&Lombroso 1971, Kutlu, et al. 2005, List, et al. 1958, Penfield 1929, Purves-Stewart 1927, Stutte 1963, Watanabe, et al. 1993). Patient expression frequently does not convey accompanying sadness or depressed mood as per patient or parents report (Dan&Boyd 1998, Hogan&Rao 2006, Lopez-Laso, et al. 2007, Luciano, et al. 1993, Tatum&Loddenkemper 2010).

The literature on dacrystic seizures is scarce, and therefore, this seizure type has been considered to be rare. However, an objective estimation of the occurrence of dacrystic seizures is not available. Additionally, an attempt to correlate the clinical presentation with underlying etiology has previously not been performed (Arroyo, et al. 1993, Dan&Boyd

1998, de Seze, et al. 1995, Hogan&Rao 2006, Kahane, et al. 1999, Kahane, et al. 2003, Kahane, et al. 1994, Lopez-Laso, et al. 2007, Luciano, et al. 1993, Procaccini, et al. 2006, Tatum&Loddenkemper 2010, Wakai, et al. 2002, Wang, et al. 1995).

Our study was designed to close these gaps. Our specific objectives were as follows: 1) to estimate the frequency of dacrystic seizures in electroencephalogram (EEG) long-term monitoring units of tertiary epilepsy centers, and 2) to correlate the clinical presentation of dacrystic seizures with underlying pathophysiology.

METHODS

Protocol approval.

This study was approved by the Institutional Review Boards and Ethical Committees of all participating institutions.

Study design.

We performed a retrospective descriptive study at five EEG long-term monitoring units at tertiary epilepsy referral centers.

Patients.

Participating epilepsy centers included University of South Florida, Cleveland Clinic, Northwestern University of Chicago in the United States of America as well as the Epilepsy Center at Freiburg University and Klinikum Osnabrück in Germany. The clinical charts and video-EEG reports of all patients admitted to the EEG long-term monitoring unit were retrospectively screened for the presence of the terms “dacrystic”, “crying”, “tears”, “tearful”, “sobbing”, “yelling”, “lacrimation”, “weeping”, and “grimacing”. Terms like “grimacing” or “yelling” are more commonly related to pain or fear and rage than to laughing. However, they can occur together with other ictal facial movements in dacrystic seizures. In the screening phase of the retrospective review we aimed to recognize the maximum possible number of patients. Then, all clinical charts and video-EEG reports with those terms were reviewed for data suggestive of the presence of dacrystic seizures. The original video-EEGs of patients with suspected dacrystic seizures were reviewed and analyzed by at least one experienced board-certified clinical neurophysiologist. We included only patients with: (1) stereotyped lacrimation, sobbing, grimacing, yelling or sad facial expression, (2) long-term video-EEG recordings (at least 12 hours), and (3) at least one magnetic resonance imaging (MRI) study.

Analysis of clinical features.

We reviewed the clinical charts in order to collect information on the demographical characteristics, types of seizures, seizure semiology, location of EEG abnormalities, neuroimaging features, neurological examination and development at the time of diagnosis of dacrystic seizures, treatments, and prognosis.

Analysis of EEG data.

Scalp-EEG recordings were placed according to the 10–20 international system of electrode placement. EEG recordings were continuously monitored by trained EEG technologists. We only included patients with at least 12 hours of EEG long-term monitoring tracing. For the purposes of this study, the original video-EEG recordings were retrieved and reanalyzed by at least one board-certified clinical neurophysiologist. The semiology of the episodes was described from the findings in the video-EEG tracing and this information was compared to and complemented with the information on semiology present in the clinical charts.

Analysis of neuroimaging features.

All original MRI studies were retrieved and re-analyzed. In all cases there was concordance with the original reports.

RESULTS

Nine patients (five males, four females) met our inclusion criteria and constituted our study population.

Frequency of dacrystic seizures.

Dacrystic seizures occurred in 0.06% to 0.53% of all the patients admitted to EEG long-term monitoring units depending on the specific center. Considering our patient population as a whole, the frequency of dacrystic seizures was 0.13% (Table 1).

Seizure semiology.

Only one patient (patient 8) presented with isolated dacrystic seizures without other manifestations. Accompanying seizure semiology included gelastic seizures in five patients and generalized tonic-clonic seizures in five patients (Table 2). The specific clinical presentation of dacrystic seizures in every patient is displayed in Table 3. An example of a typical dacrystic seizure is shown in video (Supplementary material: video).

Neuroimaging features.

Magnetic resonance imaging was abnormal in all patients, and lesions included hypothalamic hamartomas (five patients, one of them with an associated malformation of cortical development), mesial temporal sclerosis (three patients), and glioblastoma (one patient) (Table 2 and Figure 1).

Correlation of neuroimaging features and clinical presentation.

All five patients with gelastic seizures had a hypothalamic hamartoma. In four patients without gelastic seizures all lesions were cortical and three of them were located in the temporal lobe (Table 2).

EEG features.

The ictal EEG discharges were localizing most frequently to the temporal region, especially in patients with cortical lesions (Table 2 and Figure 2).

Management of epilepsy.

All patients had difficult to control epilepsy as demonstrated by the following features: (1) at least three different antiepileptic drugs were used in each patient, (2) only two patients were controlled exclusively with antiepileptic drugs, (3) six patients were considered for epilepsy surgery, and three patients underwent a surgical, radiosurgical or radioablative procedure [specific technique specified in (Schulze-Bonhage, et al. 2008)] primarily to control the epileptic syndrome (Table 2).

Outcome.

With antiepileptic drugs as the only therapy, two patients achieved seizure freedom (patients 3 and 8) and in one patient seizure frequency did not change (patient 6) on most recent follow-up (Table 2). Three patients were or are being considered for epilepsy surgery (patients 1, 2 and 9) on last follow-up. Seizure control did not improve after radiosurgery with the radioactive element I_{125} in patient 5. Patient 4 had a moderately good seizure control after treatment with I_{125} . Patient 7 remains seizure free three years after epilepsy surgery (Table 2).

DISCUSSION

Our study expands the available literature on dacrytic seizures by providing additional frequency data, semiological features, and pathophysiological relationships. We estimated the frequency of patients with dacrytic seizures in EEG long-term monitoring units of tertiary referral epilepsy centers being around 0.13% with a range of 0.06% to 0.53% (Table 1). In all cases in our series, when gelastic seizures accompanied the dacrytic seizures the underlying lesion was a hypothalamic hamartoma and when gelastic seizures did not occur the underlying etiology was of cortical (especially temporal) origin.

Frequency of dacrytic seizures.

Dacrytic seizures have been scarcely reported in literature and, therefore, they were considered to be very rare. The first description of crying seizures dates back to 1906 when Zilgien described “spasmodic crying” in a patient who finally died of luetic pachymeningitis (Zilgien 1906). Since then, the descriptions of paroxysmal and stereotyped crying were mostly limited to infrequent individual case reports and occasional descriptions of ictal crying in series of patients with other types of seizures (Breningstall 1985, Dreyer&Wehmeyer 1977, Kugoh, et al. 1988, Kutlu, et al. 2005, Oehl, et al. 2010, Offen, et al. 1976, Sethi&Rao 1976, Sher&Brown 1976, Williams, et al. 1978). A pubmed search with the search terms ‘dacrytic’, ‘crying seizures’, ‘ictal lacrimation’, ‘ictal weeping’, ‘ictal tears’ and review of citation records showed that the number of patients available in literature since 1990 with dacrytic seizures and with information on at least one brain MRI is 22 (Table 4) (Arroyo, et al. 1993, Dan&Boyd 1998, de Seze, et al. 1995, Hogan&Rao 2006, Kahane, et al. 1999, Kahane, et al. 2003, Kahane, et al. 1994, Lopez-Laso, et al. 2007, Luciano, et al. 1993, Procaccini, et al. 2006, Tatum&Loddenkemper 2010, Wakai, et al. 2002, Wang, et al. 1995). Additionally, similar to our findings, Bergen & Ristanovic reported only one case of ictal weeping out of more than 200 video-EEG tracings, leading to an approximate frequency of less than 0.5% (Bergen&Ristanovic 1993). Therefore, the

frequency of dacrystic seizures was considered to be very low. However, specific studies addressing its frequency were not available. Our data confirms that dacrystic seizures are rare and provides a numerical estimation of their frequency and clinical significance. We found a wide range of occurrence of dacrystic seizures: from 0.06% to 0.53% depending on the specific center. The frequencies found in the largest series (0.06%, 0.09%, 0.17%) or in the study population as a whole (0.13%) probably reflect better the true frequency than the higher frequencies found in smaller series (0.36%, 0.53%).

Dacrystic seizures rarely occur as an isolated clinical feature.

Dacrystic seizures frequently appear as a component of more complex seizure semiology. Gelastic seizures are the most common accompanying clinical manifestations (Hogan&Rao 2006, Lopez-Laso, et al. 2007, Luciano, et al. 1993, Tatum&Loddenkemper 2010, Wakai, et al. 2002). As reported in previous publications, dacrystic seizures in combination with gelastic seizures tend to occur in younger patients while isolated dacrystic seizures occur more frequently in older patients (Tables 4 & 5). Dacrystic seizures have been described in patients with very limited accompanying semiology like isolated staring, eye deviation, or facial automatisms (Dan&Boyd 1998, Luciano, et al. 1993). Our series adds to the previous literature by describing a patient (patient 8) with dacrystic seizures as the only clinical semiology.

Clinico-pathological correlation.

In our series, all five patients with dacrystic and gelastic seizures had a hypothalamic hamartoma and all patients with dacrystic seizures without gelastic seizures had an underlying cortical lesion. This supports available literature indicating that the presence of gelastic seizures was highly associated with an underlying hypothalamic hamartoma (Parvizi, et al. 2011). We pooled all the patients reported in the literature with dacrystic seizures since 1990 and with available information on at least one brain MRI to increase patient numbers. When added to our population of patients we found 15 patients with dacrystic and gelastic seizures. Of those, 14 patients (i.e. 93.3%) had a hypothalamic hamartoma, and one patient had a right parasagittal frontal cavernous angioma. All 14 patients with hypothalamic hamartoma had gelastic seizures. In the group of fifteen patients with dacrystic seizures without gelastic seizures we found no underlying lesion in two patients, white matter lesions in one patient, and a cortical lesion in 12 cases. Cortical lesions were located in the anterior temporal lobe in eight patients (two patients had a tumor and six had mesial temporal sclerosis) (Tables 4 & 5). Adding our data to those in previous reports, we found that patients with dacrystic and gelastic seizures have very frequent hypothalamic hamartomas (14 out of 15 patients, 93,3%) while those patients with dacrystic seizures without gelastic seizures have a relatively high rate of mesial temporal sclerosis (6 out of 15, 40%, see Tables 4 & 5). Hypothalamic hamartoma is known to be the pathologic hallmark of gelastic seizures (Striano, et al. 2009), but our analysis adds to previous literature demonstrating that patients with dacrystic seizures without gelastic seizures also frequently present with temporal lobe pathology, especially mesial temporal sclerosis (Tables 4 & 5).

Within the patient population of our tertiary epilepsy referral centers, gelastic seizures (without associated dacrystic seizures) were five times more frequent than dacrystic seizures, with a range from 25:1 in the center with the largest sample to 1:1 in the center with the smallest population. Based on this review we estimated a frequency of 0.64% of patients with gelastic seizures in our population. The large range of frequencies may have been related to selection bias: (1) centers with neurosurgical focus have a higher proportion of patients with hypothalamic hamartomas, and (2) centers with a higher proportion of children in their patient population will have overrepresentation of gelastic, but not dacrystic seizures.

Regarding the underlying pathophysiology, dacrystic seizures in patients with hypothalamic lesions could be secondary to the activation of excitable neurons in the autonomic hypothalamic centers. In contrast, dacrystic seizures in patients with cortical lesions may be related to an inhibition of actively seizing regulatory cortex leading to an autonomic release phenomenon (non-truly ictal) of the hypothalamic centers. The latter aspect has been recently supported in a patient with dacrystic seizures that occurred both during cortical discharges in the left temporal lobe and during functional inactivation of the left cortex during the Wada test. (Tatum&Loddenkemper 2010). Therefore, the probable symptomatogenic zone for dacrystic seizures is located in the hypothalamus. The mechanism of symptoms can be either direct irritation from a local tumor or a release phenomenon because of the loss of inhibition by regulatory cortex. Although it would have been interesting to correlate the presence or absence of emotional expression during seizures to an underlying pathology or localization, our data and the data in previous reports are insufficient to answer this question.

Relevance to clinical practice.

When first evaluating a patient with dacrystic seizures, the presence or absence of gelastic seizures may be able to guide the following work-up. If gelastic seizures have occurred, a hypothalamic hamartoma is the likely etiology of dacrystic seizures and the patient's autonomic function should be thoroughly studied. If gelastic seizures have not occurred, a cerebral cortical lesion, especially in the temporal lobe, should be the first option in the differential diagnosis.

Management and prognosis.

Previous literature shows that management of patients with dacrystic seizures is often challenging. Many patients are classified as refractory to medical treatment and considered for epilepsy surgery, and the overall prognosis of reported cases is frequently poor (Hogan&Rao 2006, Lopez-Laso, et al. 2007, Luciano, et al. 1993, Tatum&Loddenkemper 2010). This likely reflects the underlying etiology of the dacrystic seizures as most patients have an underlying structural brain lesion (Hogan&Rao 2006, Lopez-Laso, et al. 2007, Luciano, et al. 1993, Tatum&Loddenkemper 2010). It is unknown whether dacrystic seizures occur in patients with milder epilepsy syndromes and better prognosis and are not frequently reported (publication bias), although our series of monitored patients suggests otherwise. Nevertheless, we cannot rule out referral bias at our centers. Our data add to previous literature suggesting that patients with dacrystic seizures (1) have a high frequency of

structural brain lesions, (2) have a difficult to manage epilepsy with frequent need for epilepsy surgery and a poor prognosis.

Strengths and limitations.

Our study must be interpreted in the clinical context of data acquisition. Retrospective review of patients is associated with all the inherent bias of this type of study. We have tried to overcome these biases by: 1) collecting the information in a systematic manner, 2) retrieving and re-analysing the original EEG tracings and the original MRIs.

As patients are not always aware of ictal crying, and not all video-EEG recordings are sensitive enough to detect ictal crying, we may have overlooked patients with dacrycystic seizures in our centers. Our search terms were considered appropriate to detect patients with ictal crying, but they could have missed some patients during the searching process. Additionally, our search was performed in a population of patients referred for Video-EEG long-term monitoring at tertiary epilepsy centers. Our study population is therefore inherently biased because difficult to control seizures are likely over-represented. It is likely that dacrycystic seizures are particularly difficult to control and that they are therefore overrepresented in our study population. However, we cannot rule out that milder dacrycystic seizures are overlooked or potentially never referred to epilepsy referral centers, possibly leaving room for underrepresentation of dacrycystic seizures in our study population. Therefore, the frequency of dacrycystic seizures could have been both under- and over-estimated with our approach. However, the symptom “crying” is nonspecific and its stereotypical ictal nature can be difficult to ascertain in many cases. In order to confirm the ictal nature of the symptom “crying” in our patients, we had to review the original video recording to ascertain its stereotypical nature and we had to review the EEG tracing to confirm its occurrence during a seizure. An alternative approach of our study could have diagnosed patients based on their reported clinical presentation without considering their EEG correlate, but may have likely included many patients with non-epileptic crying or crying in response to clinical seizure manifestations. Only a prospective study with a strict definition of what dacrycystic manifestations should be and EEG confirmation of the ictal nature of the dacrycystic manifestations will provide a reliable estimation of the occurrence of dacrycystic seizures. Despite limitations, our study may therefore provide a more accurate estimate of dacrycystic seizures due to confirmation by Video EEG.

Our method of identifying patients from EEG long-term monitoring likely selected patients with a particularly difficult to control epilepsy, and a high rate of underlying structural brain lesions, epilepsy refractoriness and poor outcome could, in part, reflect this bias. Therefore, no conclusions can be drawn regarding response to pharmacological treatment and regarding outcome of dacrycystic seizures. Potential patients with easy-to-control dacrycystic seizures will be less likely to be admitted to a Video EEG monitoring unit and potentially more favorable prognosis may be underrepresented in this study. However, as stated above, previous literature consistently shows that patients with dacrycystic seizures frequently have an underlying brain structural lesion, have a difficult to manage epilepsy, and poor prognosis.

CONCLUSIONS

We expand the literature on dacrystic seizures by providing additional information on frequency, semiology and pathophysiology. Dacrystic seizures are rare in EEG long-term monitoring units of tertiary epilepsy referral centers. They are frequently accompanied by other clinical features, especially gelastic seizures. When gelastic seizures are present, hypothalamic hamartoma is the most common etiology. When gelastic seizures are not present, a cortical lesion, especially of temporal localization, is the most frequent underlying cause. Our data add to previous literature reporting that patients with dacrystic seizures generally have an underlying brain structural lesion, are frequently refractory to antiepileptic drugs, and often have a poor prognosis for seizure control.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Tobias Loddenkemper serves on the Laboratory Accreditation Board for Long Term (Epilepsy and ICU) Monitoring (ABRET), as Associate Editor for Seizure, the European Journal of Epilepsy, and on the Council of the American Clinical Neurophysiology Society (ACNS), performs Video EEG longterm monitoring, EEGs, and other electrophysiological studies at Children’s Hospital Boston and bills for these procedures, receives support from NIH/NINDS, is supported by a Career Development Fellowship Award from Harvard Medical School and Children’s Hospital Boston, by the Program for Quality and Safety at Children’s Hospital Boston, receives funding from the Epilepsy Foundation of America (EF-213583 & EF-213882), and from the Center for Integration of Medicine & Innovative Technology (CIMIT).

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Figure 1. Neuroimaging findings in patients with dacrytic seizures.

A. Sagittal T1-weighted sequence: hypothalamic hamartoma and malformation of cortical development in patient 6. **B.** Axial T1-weighted sequence: hypothalamic hamartoma and malformation of cortical development in patient 6. **C.** Coronal T1-weighted sequence: malformation of cortical development in patient 6. **D.** Sagittal T1-weighted sequence: hypothalamic hamartoma in patient 4. **E.** Axial T2-FLAIR sequence: mesial temporal sclerosis of left predominance in patient 8. **F.** Coronal T2-FLAIR sequence: mesial temporal sclerosis of left predominance in patient 9.

Figure 2. Electroencephalographic correlate of dacrytic seizures.

A. EEG onset of a dacrytic seizure in patient 4: stereotypical discharges best seen in the right sphenoidal channels. **B.** EEG onset of a dacrytic seizure in patient 9: build-up of left centro-temporal slow waves.

Table 1.

Estimated frequency of patients with dacrystic seizures among all patients admitted for EEG long-term monitoring.

	Study period (years)	Patients admitted for EEG long-term monitoring	Patients with dacrystic seizures	Estimated frequency
Cleveland Clinic	6	3153	2	0.06%
Klinikum Osnabrück	4	188	1	0.53%
Freiburg University	11	1789	3	0.17%
University of South Florida	3	1164	1	0.09%
Northwestern University at Chicago	2	557	2	0.36%
Overall study population	Not applicable	6851	9	0.13%

Table 2.

Demographic and clinical features of our patient population.

P	Age at seizure onset (years)	Handedness/gender	Seizure types	Location of EEG abnormalities	Neuroimaging	Neurologic examination	Development	TR	Age/clinical features at last follow-up
1	0.08	R/M	GS DS	R>L T-O	HH (MRI)	Macrocephaly. Rest normal.	Deficits in attention, behavior, socialization, language and learning.	TPM, TGB, PB, CLZ, CBZ, LTG, GBP, VPA, PHT (not effective)	4 years Intractable epilepsy Surgery/VNS /KD candidate
2	0.2	R/M	GS DS	Generalized	HH (MRI)	Normal	DD	KD, PHT, CLZ, LTG, CBZ, GBP, VPA, TPM, PRM (not effective)	10 years Intractable epilepsy Surgery/VNS candidate
3	34	R/M	GTC ASE DS	L F-T	L F glioblastoma (MRI)	Right-sided hemiparesis, dysarthria, mild-moderate non-fluent aphasia	Normal	Chemotherapy, radiotherapy, LZP, LEV (not effective) PHT (effective)	34.3 Seizure free on PHT
4	10	R/M	GS DS DYS GTC	R F-T	R-sided HH (MRI) Hypometabolism in right thalamic area (PET) Low blood flow in temporobasal areas bilaterally (interictal SPECT)	Normal	DD	PHT, VPA (not effective) Iodid 125 at age 15	24 Rare seizures
5	9	R/F	GS DS DYS GTC Heart rate	B F-T	L-sided HH (MRI) Hypometabolism in left parietal and	Normal	DD	PHT, VPA (not effective) Radiosurgery at age	23 No significant seizure reduction

6	1	R/F	GS Automotor DS	R C	bilateral temporal (PET)	Normal	DD	16 Iodid 125 at age 18	9 No significant seizure reduction
7	0.8	R/F	First seizure: GTC Following seizures: aura and DS Left carotid methohexit al anesthesia triggered typical DS	L T	L MTS (MRI) L T hypometabolis m (PET) Increased L T cerebral blood flow (ictal SPECT)	Normal	Normal	PB, AZM, TPM, LEV, CBZ (all drugs were ineffective in producing seizure freedom) Left amygdalo - hypocam ectomy at age 32	35 years Following amygdalo-hypocampe ctomy: seizure freedom on tapering doses of CBZ Quality of life has largely improved. She is driving and has entered a complete college education
8	70	R/M	DS	L T	MTS L>R (MRI) L mesial temporal hypometabolis m (PET) Increased left mesial temporal cerebral blood flow (SPECT)	Normal	Normal	PHT, LEV, OXC (not effective) TPM (effective)	73.3 Seizure free
9	9	R/F	First seizure: GTC Following seizures: Auras and loss of awareness DS	B T	MTS L>R (MRI)	Normal	Normal	LTG, GBP, CBZ, LCS, TPM, CBZ (not effective)	38 years Intractable, not a surgical candidate. Failed Wada test.

Legend: ASE: aphasic status epilepticus. AZM: acetazolamide. B: bilateral. C: central. CBZ: carbamazepine. CLZ: clonazepam. DD: developmental delay. DS: dacrystic seizure. DYS: dyscognitive seizures. F: frontal. GBP: gabapentin. GS: gelastic seizure. GTC: generalized tonic-clonic seizure. HH: hypothalamic hamartoma. KD: ketogenic diet. L: left. LCS: lacosamide. LEV: levetiracetam. LTG: lamotrigine. LTP: lorazepam. MRI: magnetic resonance imaging. MTS: mesial temporal sclerosis. O: occipital. P: patient. PB: phenobarbital. PHT: phenytoin. PRM: primidone. R: right. T: temporal. TGB: tiagabine. TPM: topiramate. TR: treatment. VNS: vagal nerve stimulator. VPA: valproate.

Table 3.

Specific clinical presentation of the dacrystic seizures in our patients.

Patient	Lacrimation	Grimacing	Sobbing	Sad facial expression	Yelling	Sadness reported by parents	Typical duration of the dacrystic component	Typical frequency of seizures	Presence of postictal symptoms
1	N	Y	N	Y	N	N	20s	10/day	N
2	N	Y	N	Y	Y	N	10–30s	4/day	Confusion
3	N	Y	N	Y	N	Y	30–60s	2/day	N
4	Y	Y	N	N	N	N	5s	2–3/week	N
5	Y	N	N	Y	N	N	15s	2/day	Anomia and aphasia
6	N	Y	Y	Y	N	Developmentally noncommunicative	15s	3/day	N
7	N	N	N	Y	N	N	20s	1–2/week	Brief fatigue, confusion and aphasia
8	N	Y	N	Y	N	Immediately after but not during the event	10–20s	5–20/day	Disorientation and depressed mood
9	N	Y	Y	Y	Y	N	1 min	2–4/month	N

Legend: N: no. Y: yes.

Table 4.
Previous literature on dacrycystic seizures.

We have excluded studies: 1) performed before 1990 or 2) that do not provide information on at least one brain MRI.

Author & year	Number of patients	Age(years)/sex	Gelastic seizures	MRI	EEG localization (superficial EEG)	Surgery (pathology)
(Arroyo, et al. 1993)	1	35/F	Y	R parasagittal F cavernous angioma	L FT	MR (cavernous hemangioma)
(Luciano, et al. 1993)	7	34/M	N	Increased signal R ATL	R MT	R ATL (neurocytoma)
		61/M	N	Mixed density lesion in R ATL	B MT	R ATL (astrocytoma/oligodendroglioma)
		35/F	N	Normal	R MT	R ATL (MTS)
		25/F	N	R F cavernous hemangioma	B FT	LT (cavernous hemangioma)
		32/F	N	R hemiatrophy	R ATL	R ATL (normal)
		32/M	N	Normal	MI FC	No
		48/F	N	Normal	R MT	No
(Kahane, et al. 1994)	1	20/F	Y	HH	R centropetal	No
(de Seze, et al. 1995)	1	33/M	N	L MTS SPECT: bilateral frontal hypometabolism	Right temporal	No
(Wang, et al. 1995)	1	66/M	N	Focal ischemic foci in the posterior limb of the right internal capsule	Right temporal	No
(Dan&Boyd 1998)	1	0.5/M	N	Subependymal nodules and cortical hamartomas	No ictal EEG	No
(Kahane, et al. 1999, Kahane, et al. 2003)	2	19/F	Y	HH	R FCT	MR
		27/M	Y	HH	NI	No
(Wakai, et al. 2002)	1	4/F	Y	HH	Left central	No
(Hogan& Rao 2006)	1	28/M	N	Normal (PET hypometabolism in the L ATL)	L ATL	L ATL (MTS)
(Procaccini, et al. 2006)	4	0.08/M	Y	HH	NI	HH dysconnection
		0.5/M	Y	HH	NI	HH dysconnection
		6/M	Y	HH	NI	HH dysconnection
		1/M	Y	HH	NI	HH dysconnection
(Lopez-Laso, et al. 2007)	1	0.08/F	Y	HH	Normal EEG	MR (HH)
(Tatum&Loddenkemper 2010)	1	0.8/F	N	L MTS	L FT	L AH (MTS)
Blumberg et al	9	0.08/M	Y	HH	R T	Surgical candidate
		0.2/R	Y	HH	Generalized spike-wave	Surgical candidate
		34/R	N	L frontal glioblastoma	L FT	No
		10/R	Y	HH	R FT	No. Radiotherapy with I ₁₂₅

Author & year	Number of patients	Age(years) /sex	Gelastc seizures	MRI	EEG localization (superficial EEG)	Surgery (pathology)
		9/R	Y	HH	B FT	Radiosurgery and radiotherapy with I ₁₂₅
		1/R	Y	HH and complex gray-matter malformation	R C	No
		70/R	N	L MTS	L T	No
		9/R	N	L MTS	L T	No

AH: amygdalo-hippocampectomy. ATL: anterior temporal lobe. ATLT: anterior temporal lobectomy. B: bilateral. F: female/frontal. FC: fronto-central. FT: fronto-temporal. HH: hypothalamic hamartoma. L: left. LT: lesionectomy. M: male. MI: midline. MR: mass resection. MRI: magnetic resonance imaging. MT: mesial temporal. MTS: mesial temporal sclerosis. N: no. NI: no information. R: right. TL: temporal lobectomy. Y: yes.

Table 5.

Underlying etiology in our population of patients.

	Hypothalamic hamartoma	Abnormal white matter	Cortical lesion	No lesion
Patients with dacrystic and gelastic seizures (N=15)	N=14 *	N=0	N=2 *	N=0
Patients with dacrystic without gelastic seizures N=15	N=0	N=1	N=12	N=2

N=number of patients in each category

* Patient 6 in our series had hypothalamic hamartoma and malformation of cortical development.