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Coprolalia as a manifestation of epileptic seizures

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ABSTRACT

Objective: The aim of this study was to investigate the lateralizing and localizing value of ictal coprolalia and brain areas involved in its production.
Methods: A retrospective search for patients manifesting ictal coprolalia was conducted in our EMU database. Continuous video-EEG recordings were reviewed, and EEG activity before and during coprolalia was analyzed using independent component analysis (ICA) technique and was compared to the seizures without coprolalia among the same patients.
Results: Nine patients were evaluated (five women), eight with intracranial video-EEG recordings (icVEEG). Four had frontal or temporal lesions, and five had normal MRIs. Six patients showed impairment in the language functions and five in the frontal executive tasks.
Two hundred six seizures were reviewed (60.7% from icVEEG). Ictal coprolalia occurred in 46.6% of them, always associated with limbic auras or automatisms. They arose from the nondominant hemisphere in five patients, dominant hemisphere in three, and independently from the right and left hippocampus–parahippocampus in one. Electroencephalographic activity always involved orbitofrontal and/or mesial temporal regions of the nondominant hemisphere when coprolalia occurred.
Independent component analysis of 31 seizures in seven patients showed a higher number of independent components in the nondominant hippocampus–parahippocampus before and during coprolalia and in the dominant lateral temporal region in those seizures without coprolalia ($p = 0.009$). Five patients underwent surgery, and all five had an ILAE class 1 outcome.
Significance: Ictal coprolalia occurs in both males and females with temporal or orbitofrontal epilepsy and has a limited lateralizing value to the nondominant hemisphere but can be triggered by seizures from either hemisphere. It involves activation of the paralimbic temporal–orbitofrontal network.

1. Introduction

Seizure semiology is crucial in the presurgical evaluation of patients with epilepsy. In this regard, ictal speech lateralizes to the nondominant hemisphere for language and ictal dysphasia to the dominant one in temporal lobe epilepsy (TLE). Conversely, there are contradictory data on localization of ictal vocalizations [1–6].

Coprolalia is the involuntary utterance of socially unacceptable words, including obscenities, or ethnic, racial, or religious slurs [7]. It is a well-known symptom of Tourette syndrome (TS) [8] and has also been described in dementia, brain tumors, and other neurological diseases [8,9]. However, it has rarely been reported in seizures [10].

Independent component analysis (ICA) is a mathematical algorithm that is able to decompose the combination of neural signals producing an EEG recording to yield their individual sources [11,12]. Independent component analysis should work just as well for separating invasive EEG signal mixtures, providing a clinically feasible way to separate independent components (ICs), proximal and distal in origin, to better depict their distribution [11].

The aim of the present study was to elucidate the localizing and lateralizing value of ictal coprolalia and the brain regions involved in its production. A detailed description of the ictal semiology and EEG activity and its ICs at the time of coprolalia is provided for a series of patients. An Illustrative case in the online version at http://dx.doi.org/10.1016/j.yebeh.2016.04.040. is described.

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2. Methods

2.1. Patients and presurgical evaluation

A retrospective search for patients manifesting with ictal coprolalia was made in the database of our epilepsy monitoring unit (EMU) among patients admitted from July 2008 to November 2014.

Coprolalia was defined as the involuntary utterance of obscene words or socially inappropriate and derogatory remarks. Obscenities are defined as utterances describing sexual acts, body function or elimination acts, organs of reproduction and sexual anatomy, references to animals, or racial epithets. Profane utterances were defined as those conveying a religious connotation [8].

Evidence of associated epilepsy was defined as an unequivocal ictal pattern.

Patients underwent continuous video-EEG telemetry monitoring using standard scalp electrodes followed by implanted subdural or depth EEG electrodes. Scalp EEG, based on the extended 10–20 International System of electrode placement plus two mandibular notch electrodes, was recorded in accordance with the Canadian Society of Clinical Neurophysiology guidelines [13,14]. Intracranial video-EEG recordings (icVEEG) were monitored using 128-channel XLTEK equipment (Oakville, ON) at sampling rates of 1000 Hz.

Strategy of electrode positioning was established in each patient based on hypotheses about epileptogenic zone localization. Subdural strip electrodes were implanted through burr holes. Our technique for this has been described elsewhere [15,16]. For temporal lobe coverage, two or three lines of eight contacts each were inserted via a posterior temporal burr hole to cover the mesial, inferior, and/or lateral surfaces of the temporal lobe. Patients four and eight had additional subdural strip and depth electrode coverage of the parietal and frontal lobes, respectively. Patients six and nine were studied with depth electrodes covering the temporal, insular, cingulate, and frontal regions. See Table l for further details.

Language lateralization was evaluated by the Fused Rhyme Dichotic Words Test [17,18], functional MRI, and Wada tests. In patients two, three, and seven, language tests could not be completed or were invalid because of too many errors. In these cases, lateralization was estimated from handedness and features of their epilepsy.

Neuropsychological assessments were performed prior to surgery according to a standard protocol used at our center. Psychological profiles were obtained from a clinical psychological evaluation, including the Minnesota Multiphasic Personality Inventory Version 2 [19].

Clinical data were systematically collected through chart review, and the seizure outcomes were classified using ILAE classification [20]. Seizures were explored by trained nursing staff.

Written informed consent was obtained from all the participants in the study, including authorization for disclosure of any recognizable persons in photographs, videos, or other information published. The study was approved by our institutional research ethics board.

2.2. Independent component analysis

Independent component analysis was performed using EEGLAB toolbox [21] in MATLAB software (Mathworks, Natick, MA). Initial processing involved bandpass filtering the data between 1 and 70 Hz in order to remove artifact contamination and other baseline wandering. The JADE algorithm was selected to apply ICA [22].

In each case, ICA was applied to the recordings of all channels resulting in the same number of ICs as the number of channels. Validity of the output from ICA was confirmed by regenerating the original EEG from the ICA output using a reverse mixing process. The significant ICs were then selected visually. The corresponding location of each significant IC was detected by calculating the correlation of each IC to the signals of all channels and choosing the channels with the highest correlation values.

Independent component analysis was performed on seven patients, six with seizures recorded on icVEEG and one on scalp. Independent component analysis could not be performed on the other two patients (patients three and four) because of technical problems with the storage of their seizures.

Seizures were split into the following sections for analysis:

For those seizures with coprolalia: a) Precoprolalia, defined as the period between the EEG onset of the seizure and the beginning of ictal coprolalia; b) coprolalia, defined as the period during which the patient manifested coprolalia; and c) postcoprolalia, defined as the period between the end of coprolalia and the electrographic ending of the seizure.

For seizures without coprolalia: a) Onset, the period between the EEG seizure onset and the spreading of the activity; b) spreading, the period between the start and the end of the EEG activity spread; and c) end, the period from the end of spreading to the end of the seizure.

For an approximate matching, precoprolalia and coprolalia periods were compared with the onset and spreading periods.

In patients with seizures with coprolalia from more than one seizure focus (patient seven), seizures from each focus were analyzed separately.

The localization of the intracranial EEG electrodes involved in each IC was represented in maps extrapolated from CT–MRI superimposition images generated by Atamai software [23] (Fig. 1).

2.3. Statistics

Analyses were performed with the SPSS statistical package (Chicago, IL, USA, version 17.0). For continuous variables that were normally distributed, values were expressed as means ± SD, and statistical significance for intergroup differences was assessed using independent samples Student's t-test. A p value of <0.05 was considered statistically significant. For variables not normally distributed, values were expressed as medians (range).

3. Results

3.1. Baseline characteristics

Among 1381 patients evaluated in our epilepsy monitoring unit between July 2008 to November 2014, nine cases (five women) with ictal coprolalia were identified (0.65%). The baseline characteristics of the study population and the electroclinical description of their seizures are summarized in Table 1.

All patients suffered focal dyscognitive seizures, and seven had bilateral convulsive seizures. One patient also had psychogenic nonepileptic seizures. All except two patients had seizures with coprolalia arising from NREM sleep, but they were the predominant type only in patient nine.

Three patients had an underlying frontal or temporal ganglioglioma, and one had a focal cortical dysplasia in the left temporal lobe. Five patients had normal anatomical MRI with negative autoimmune workup, except for elevated antithyroid peroxidase (127 IU/ml, normal value ≤ 34 IU/ml) and antithyroglobulin antibodies (330 IU/ml, normal value ≤ 115 IU/ml) with normal thyroid hormone levels in one (patient seven).

All but one patient underwent icVEEG. Two hundred six seizures (range: 3–48 per patient) were evaluated, 60.7% of these with icVEEG.

Five patients underwent surgery. All became free of disabling seizures (ILAE classes 1–2) for a minimum of one year.

3.2. Neuropsychological and psychiatric evaluation

All except patient three completed the neuropsychological evaluation. They all displayed some degree of impairment in one or more of the four neuropsychological domains (language, verbal memory, visual memory, and frontal lobe executive skills). Specifically, six showed a