

Epistaxis During a Generalized Seizure Leading to an Atypical Ictal SPECT Finding at the Skull Base

Lauro Wichert-Ana^{***}, Emerson Henklain Ferruzzi^{*}, Veriano Alexandre Jr.^{*}, Tonicarlo Rodrigues Velasco^{*}, Marino Muxfeldt Bianchin^{*}, Vera Cristina Terra-Bustamante^{*}, Mery Kato^{**}, Antonio Carlos Santos^{***}, Paulo Mazzoncini de Azevedo-Marques^{***}, Lucas Ferrari de Oliveira^{***}, Américo Ceiki Sakamoto^{*}

Ribeirão Preto School of Medicine, University of São Paulo, USP

ABSTRACT

Introduction: Generalized tonic-clonic seizures (GTCS) are among the most dramatic types of epileptic seizures and may be accompanied by rising blood pressure and pulse rate, physical injuries from falling, muscular convulsions, tongue biting, or aspiration pneumonia. Epistaxis is an uncommon complication of generalized seizures and investigations should exclude local or systemic disorders. **Objective:** We aim to report a 29-year-old male patient with medically intractable right temporal lobe epilepsy whose ictal SPECT showed a conspicuous high extracerebral accumulation of the tracer at the skull base. **Methods:** The tracer ^{99m}Tc-ECD was injected during a GTCS complicated by simultaneous epistaxis during a long term video-electroencephalographic monitoring. **Results:** Initially, SPECT images showed an unexpected hot spot at the skull base suggesting pharyngeal or pituitary tumors. Clinical history disclosed chronic sinusitis and rare episodes of epistaxis. White and red cells blood count, platelet count, serum biochemistry, coagulation tests, and rest arterial blood pressure were normal. Computed tomography and MRI excluded sinusoidal expansive or vascular lesions, head trauma, fractures or acute infections. Subtracted SPECT disclosed a focal high concentration of the radiotracer within the left sphenoid sinus, probably related to the nose bleeding. **Conclusion:** This is a singular case of a brain SPECT artifact secondary to a nasal bleeding during a generalized seizure that was misinterpreted as neoplastic disease. Also, this case raises concerns about the pathophysiological relationship among epileptic seizures, nasal bleedings and chronic sinusitis.

Key words: epilepsy, epistaxis, generalized seizure, SISCOM, SPECT.

RESUMO

Epístaxe durante crise generalizada resultando em um achado atípico na base do crânio ao SPECT crítico

Introdução: As crises generalizadas tônico-clônicas (CGTC) constituem-se em formas dramáticas de crises epilépticas e podem acompanhar-se de aumento da pressão arterial e da frequência cardíaca, traumas decorrentes de quedas, abalos musculares, mordedura de língua e pneumonias aspirativas. A epístaxe é uma complicação incomum e investigações médicas devem excluir distúrbios locais ou sistêmicos. **Objetivo:** Relatar o caso de um paciente de 29 anos de idade com epilepsia do lobo temporal direito clinicamente intratável e cujo SPECT crítico mostrou uma área de acúmulo anormal do traçador na base do crânio. **Método:** O traçador ^{99m}Tc-ECD foi injetado durante uma CGTC complicada por simultânea epístaxe na narina esquerda durante a monitorização vídeo-eletroencefalográfica. **Resultados:** O SPECT crítico evidenciou área de acúmulo anormal do traçador na base do crânio sugerindo tumor de natureza neuronal ou glial e de origem faríngea ou pituitária. A história clínica evidenciou sinusite crônica e raros episódios de epístaxe. Exames hematológicos das séries branca e vermelha, contagem de plaquetas, bioquímica sérica, testes de coagulação e medidas de pressão arterial em repouso foram normais. A Tomografia Computadorizada e a Ressonância Magnética

* Department of Neurology (Epilepsy Surgery Center).

** Division of Nuclear Medicine of the Department of Internal Medicine.

*** Imaging Science and Medical Physics Center of the Department of Internal Medicine. Ribeirão Preto School of Medicine, University of São Paulo, USP, Ribeirão Preto, Brazil.

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(RM) excluíram lesões expansivas ou vasculares, trauma craniano, fraturas ou infecções agudas. A subtração baseada em voxel das imagens de SPECT crítico e intercrítico alinhada ao espaço 3D da RM evidenciou uma alta concentração do traçador no seio esfenoidal esquerdo. **Conclusão:** Este é um caso singular de um artefato ao SPECT crítico secundário ao sangramento nasal durante uma crise epiléptica e que foi inicialmente interpretado como doença neoplásica. Este caso também indaga sobre o possível relacionamento fisiopatológico entre crises epilépticas, sangramentos nasais e sinusite crônica.

Unitermos: epilepsia, epistaxe, crises generalizadas, SISCOM, SPECT.

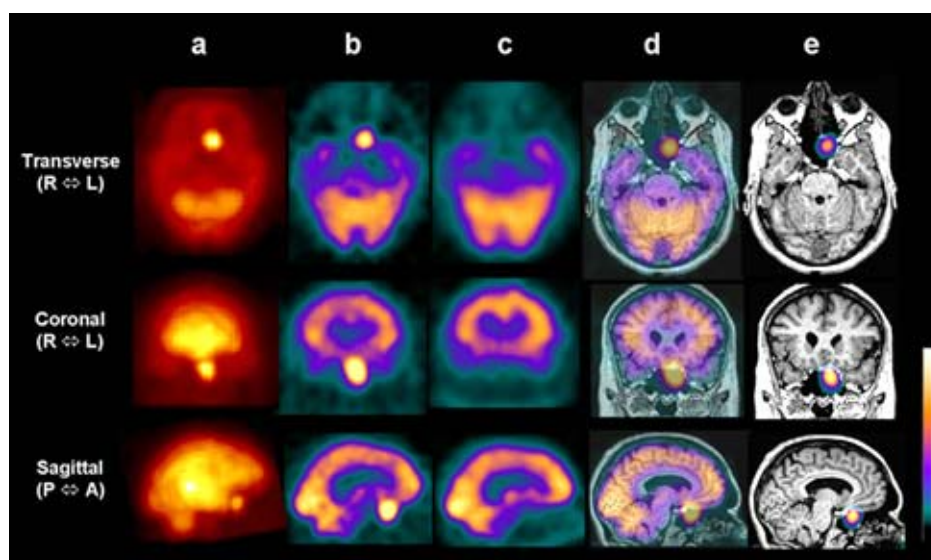
INTRODUCTION

Generalized tonic-clonic seizures (GTCS) are among the most dramatic types of epileptic seizures. They may begin abruptly or may be preceded by simple or complex partial seizures, evolving with loss of consciousness, generalized muscular contraction, forced continued expiration, rhythmic jerking lasting few seconds to several minutes, and rising blood pressure and pulse rate⁽¹⁾. Physical injuries from falls, muscular convulsions, tongue biting, or aspiration pneumonia may complicate GTCS⁽¹⁾. Reports of epistaxis during GTCS are rare. Some of them refer to primary nasal tuberculosis causing ictal nose bleeding⁽²⁾, while others deal with haemostatic disorders associated to antiepileptic drugs⁽³⁾.

We report here ictal SPECT findings during GTCS complicated by epistaxis showing an abnormal accumulation of the radiotracer in the skull base. Subtraction ictal SPECT Co-registered to Magnetic Resonance (SISCOM) disclosed a very singular extracerebral radioactive blood leak.

CASE REPORT

A 29-years-old male with medically intractable epilepsy was referred to our Epilepsy Surgery Center for presurgical evaluation. His epilepsy started with 17 years old and he was currently taking phenytoin and clobazam. Neurological examination was normal and 1.5T Magnetic Resonance Imaging (MRI) had shown bilateral hippocampus atrophy, worst in the right side. During video-EEG monitoring (VEEG) we recorded complex partial seizures evolving to GTCS, with ictal findings consistent with right temporal lobe origin. The radiotracer injection for ictal SPECT was completed during the generalized phase of one of the seizures that was associated to nasal bleeding through the left nostril. The bleeding stopped spontaneously in the postictal period. Ictal SPECT showed an atypical and very high accumulation of the radiotracer at the skull base, suppressing the normalized visualization of the rest of brain activity, while interictal SPECT only showed mild hypoperfusion in the right temporal lobe. SISCOM improved localization and revealed that the extracerebral accumulation of the radiotracer was located within the left sphenoid sinus (see Figure).



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Figure 1. Left sphenoid sinus bleeding during generalized tonic-clonic seizure, exhibited in transverse, coronal and sagittal slices

(R: right; L: left; A: anterior; P: posterior). (a) 3D rendered volume of ictal SPECT, (b) Ictal SPECT showing abnormal accumulation of the radiotracer at the skull base, (c) Interictal SPECT shows a mild hypoperfusion in the mesial region of the right temporal lobe. (d) Co-registration of ictal SPECT with MRI and (e) Subtracted ictal-interictal SPECT pinpoint the radioactive blood volume in the left sphenoid sinus.

The patient had informed he suffered from chronic repetitive sinusitis and had previous spontaneous nose bleedings, but no recent flu, rhinitis or sinusitis symptoms. His lab tests including white and red blood cells count, platelet count, serum biochemistry and coagulation tests were all normal. Arterial blood pressure was 130/80 mmHg and heart rate at rest was 88 beats/min. Computed tomography, performed one week after, excluded sinusoidal expansive or vascular lesions, head trauma, fractures or acute infections. Patient did not undergo epilepsy surgery yet.

DISCUSSION

We report here an ictal SPECT hot spot finding localized at the skull base during a GTCS of an epileptic patient whose clinical history and structural neuroimaging excluded the possibility of neuroglial tumor. What makes it a very singular case is the occurrence of left nostril epistaxis during the injected GTCS, and a SISCOM precisely pinpointing the left sphenoid sinus as the probable bleeding site. SPECT images were initially misinterpreted as pharyngeal mass lesion, corresponded, in fact, to an extracerebral accumulation of a radioactive blood volume. Beyond the rarity of this flagrant SPECT image, this case also raised some interesting discussion points.

First, nose bleeding during epileptic seizures are rare in patients without other comorbidities. Epistaxis, petechiae, echymosis and other hemostatic signs are uncommon side effects of antiepileptic drugs⁽¹⁾. Complete blood cells and platelet counts and a prothrombin/international normalized ratio (PT/INR) assessment should be performed to exclude neutropenia or hemostasis disturbances. Our patient had normal coagulation functions and did not have other bleeding disorders except epistaxis.

The second point is the "perfusion" pattern observed in the ictal SPECT. Similar PET or SPECT findings with

different radiotracers have been observed in pharyngeal and pituitary neuroendocrine tumors or abscess^(4,5). Our clinical and laboratory investigations excluded the presence of mass lesions. Probably, either arterial or venous hemodynamic changes during the GTCS⁽¹⁾ could have led to a blood leak in a chronic inflamed or infected left sphenoid sinus, reaching through the sphenoidal recess the left nasal cavity. The focal accumulation of the radiotracer at the skull base was, in fact, an extracerebral SPECT artifact documented by SISCOM.

Finally, our patient had chronic sinusitis and we have previously observed that this condition appears to be more prevalent in epileptic patients than in controls (unpublished data). We speculate whether the sum of unperceived ictal sinus bleedings, associated with excessive mucous discharge secondary to seizures or benzodiazepines, ictal drooling or vomiting and drug-induced gingivitis with dental infections could facilitate chronic sinusitis in epileptic patients.

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Address correspondence to:

Lauro Wichert Ana
Hospital das Clínicas da Faculdade de Medicina de Ribeirão Preto
Campus Universitário
CEP 14048-900, Ribeirão Preto, SP, Brazil
Phone: (55016)602-2613 – Fax: (55016)633-0760
E-mail: lwichert@mp.fmrp.usp.br