

Case Report

An unusual presentation of dystonia and chorea from intraventricular pneumocephalus

Sean P. Polster^{1,2}, Shirlene Obuobi³, Victor J. Del Brutto³, Kenneth Avner³, Aikaterini Markopoulou³, Ricky H. Wong²

¹Department of Surgery, Section of Neurosurgery, University of Chicago, Chicago, Departments of ²Neurosurgery and ³Neurology, Northshore University Health System, Evanston, Illinois, USA

E-mail: Sean P. Polster - spolster@uchicago.edu; Shirlene Obuobi - shirlene.obuobi@uchospitals.edu; Victor J. Del Brutto - victor.delbruttoandrade@uchospitals.edu; Kenneth Avner - kenneth.minorczyk@uchospitals.edu; Aikaterini Markopoulou - amarkopoulou@northshore.org; *Ricky H. Wong - wong.ricky@gmail.com

*Corresponding author

Received: 15 June 18 Accepted: 23 August 18 Published: 21 September 18

Abstract

Background: Pneumocephalus is a common finding following intracranial procedures, typically asymptomatic and resolves within several days. However, in some cases, pneumocephalus presents with headache, encephalopathy, or symptoms of elevated intracranial pressure. Here, we present a case of iatrogenic tension pneumocephalus following endoscopic sinus surgery, presenting as abnormal involuntary movements resembling a movement disorder with choreiform movements.

Case Description: A 67-year-old previously healthy male presented with new onset chorea and dystonia associated with headache, encephalopathy, and postural instability 4 days after undergoing endoscopic sinus surgery for chronic sinusitis and nasal polyps. Computed tomography showed prominent intraventricular pneumocephalus causing enlargement of the anterior horns of both lateral ventricles with lateral displacement of the basal ganglia nuclei and a bony defect in the skull base. Neurosurgical correction of the cranial defect provided complete symptomatic resolution. Pneumocephalus as a result of an iatrogenic injury of the skull base manifesting as an acute movement disorder is a rare complication of a nasal sinus procedure. We speculate that compression of the caudate nucleus and striatum resulted in decreased pallidothalamic inhibition and thalamocortical disinhibition leading to the development of a hyperkinetic movement disorder.

Conclusion: This unusual presentation of a common procedure illustrates a neurological emergency that requires prompt recognition and timely correction.

Key Words: All movement disorders, clinical neurological examination, dystonia

Access this article online

Website:

www.surgicalneurologyint.com

DOI:

10.4103/sni.sni_190_18

Quick Response Code:

INTRODUCTION

Tension pneumocephalus resulting in caudate compression is a unique etiology of choreiform movements.

CASE DESCRIPTION

A 67-year-old man with no significant medical history presented to the emergency department (ED) with

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How to cite this article: Polster SP, Obuobi S, Brutto VJ, Avner K, Markopoulou A, Wong RH. An unusual presentation of dystonia and chorea from intraventricular pneumocephalus. *Surg Neurol Int* 2018;9:193.

<http://surgicalneurologyint.com/An-unusual-presentation-of-dystonia-and-chorea-from-intraventricular-pneumocephalus/>

a one-day course of new onset headache, chorea, postural instability, and disorientation. Four days before presentation, he underwent endoscopic sinus surgery for chronic sinusitis and nasal polyps at an outpatient sinus surgery center. The operation was reported as uneventful and he was discharged home neurologically intact. No medications were prescribed nor used without prescription by the patient. Three days after surgery, the patient developed bilateral frontal headache associated with poor movement control, inability to ambulate or operate his automobile. At arrival to the ED, he was afebrile, hemodynamically stable, and in no acute distress. On neurological examination, he was awake, but inattentive, oriented to self and place but not time. There was no nuchal rigidity and cranial nerves were intact. He was noted to have involuntary chorea-like movements of the neck and shoulders as well as dystonic posturing of his right arm and neck. His muscle strength was preserved and tone was increased in the lower extremities. Right patellar hyperreflexia, right ankle clonus, and right extensor plantar response were present. Sensory examination was unremarkable. He was unable to stand due to significant postural instability. Clear fluid rhinorrhea was noted during the exam. Computed tomography on presentation showed prominent pneumocephalus with intraventricular air causing enlargement of the anterior horns of both lateral ventricles with lateral displacement of the basal ganglia nuclei bilaterally [Figure 1a] and a bony defect at the lateral lamella and fovea ethmoidalis with formation of an encephalocele [Figure 1b and c]. Emergent endoscopy confirmed an anterior skull base defect of approximately 1.5 cm with associated encephalocele [Figure 2]. A multilayer repair was completed using synthetic inlay, an inferior turbinate-free mucosal graft onlay, and DuraSeal (Integra Life Science). His postoperative course was characterized by gradual improvement in neurological symptoms. On postoperative day 3, the patient was noted to have a normal neurological examination with radiographic resolution of the pneumocephalus and he was discharged home.

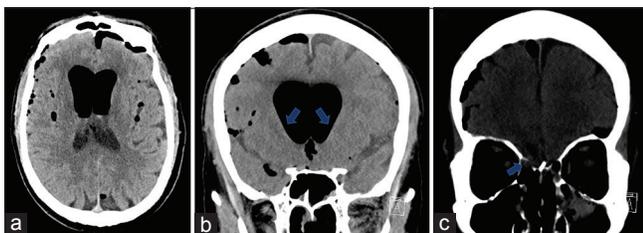


Figure 1: Nonenhanced computerized tomography scans showing in the (a) axial and (b) coronal planes, extra-axial, subarachnoid, and intraventricular air (arrows in b shows depression of the ventricular wall causing distortion of the bilateral caudate nuclei). (c) Coronal computerized tomography scan with arrow depicting bony defect caused by iatrogenic transgression of the fovea ethmoidalis

DISCUSSION

Pneumocephalus as a result of an iatrogenic injury of the skull base presenting as an acute movement disorder is a rare complication of a nasal sinus procedure. This complication resulted from the violation of the subdural and subarachnoid compartments resulting in CSF drainage and intraventricular accumulation of air under pressure. The intraventricular air caused lateral ventricle distortion that compressed and displaced the bilateral basal ganglia nuclei inferolaterally.

Pneumocephalus is a routine radiographic finding after intracranial procedures, usually asymptomatic and resolves within days. Infrequently, iatrogenic violation of the dura with subsequent CSF loss results in a “valve” phenomenon with intracranial accumulation of air that can cause mass effect on surrounding structures. A dramatic example is the “Mt Fuji sign” with bilateral frontal lobe compression. Further communication to the ventricular system and subarachnoid space can lead to intraventricular accumulation of air and progressive neurological deterioration.^[1,5,6] While headache and encephalopathy are common presentations of tension pneumocephalus, abnormal involuntary movements have not been associated with intraventricular tension pneumocephalus and to our knowledge have not been previously reported.^[7]

Historically, isolated intraventricular air was most commonly encountered after pneumoencephalography. Neurological complications of this air procedure lack a particular pattern and include headaches, alteration of consciousness, and seizures as well as abnormal pyramidal and extrapyramidal system involvement.^[8] However, with this diagnostic modality falling out of use in modern medicine, intraventricular pneumocephalus is rarely seen.

Lutjens and colleagues reported a single case of akinetic mutism and Parkinsonism associated with

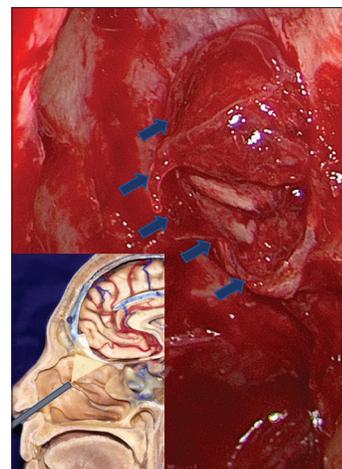


Figure 2: Endoscopic endonasal view via the right nare with arrows showing bony defect edge with dural violation and encephalocele. Insert shows perspective (The Rhoton collection)

tension pneumocephalus after decompression of a subdural hygroma where downward pressure distorted the frontal hemispheres and deep nuclei.^[4] Our case demonstrates development of focal chorea and dystonia associated with upper motor neuron signs and encephalopathy due to compression and displacement of basal ganglia nuclei by intraventricular tension pneumocephalus. We speculate that bilateral compression of the caudate nucleus and striatum by intraventricular air in the anterior horns of the lateral ventricles resulted in decreased pallidothalamic inhibition and thalamocortical disinhibition leading to the development of reversible abnormal movements. Similarly, bilateral basal ganglia injury caused by autoimmune disorders, drugs, hypoxia, metabolic disorders, or infection has been reported to present as acute chorea.^[2,3]

This presentation illustrates a neurosurgical emergency that requires prompt recognition and intervention.^[5,6] Timely correction of the anatomical defect in this individual resulted in complete recovery of neurological function. Surgeons performing endonasal procedures and physicians evaluating patients in the ED for acute onset of movement disorders should be aware of this uncommon presentation.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported

in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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