Case Report

Spontaneous pneumocephalus secondary to a cerebrospinal fluid fistula demonstrated by CT cisternography

Lien Van Liedekerke, MD\textsuperscript{a}, Said Sanoussi, MD\textsuperscript{b,}\textsuperscript{*}

\textsuperscript{a}Department of Neuroradiology, Brugmann University Hospital, Vrije Universiteit Brussel (VUB), Brussels, Belgium
\textsuperscript{b}Department of Neuroradiology, Brugmann University Hospital, Université Libre de Bruxelles (ULB), Brussels, Belgium

\textbf{ARTICLE INFO}

Article history:
Received 25 May 2020
Revised 13 July 2020
Accepted 13 July 2020

Keywords:
Pneumocephalus
Cerebrospinal fluid leak
Cisternography

\textbf{ABSTRACT}

We report a case of a middle-aged woman who presented to our emergency department with increasing headache in a nontraumatic setting. The presence of intracranial air was an unexpected finding on nonenhanced computed tomography (CT). CT and magnetic resonance imaging could not identify the origin of the bone defect responsible for pneumocephalus. CT cisternography was able to demonstrate the presence of a cerebrospinal fluid fistula resulting in pneumocephalus. This case highlights the role of CT cisternography to identify and localize small osseous defects and cerebrospinal fluid fistulas when CT and magnetic resonance imaging findings are normal.

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\textbf{Introduction}

Pneumocephalus is defined as the presence of air within the intracranial cavity. Air can be present in the epidural, subdural, subarachnoid, intraventricular, or intraparenchymal compartments [1–4].

In the vast majority of cases, pneumocephalus is asymptomatic. Persistent or tension pneumocephalus, when intracranial air is trapped resulting in mass effect, can result in headache and signs and symptoms of increased intracranial pressure [2,3,5].

The main cause of pneumocephalus is trauma resulting in fractures of air sinuses or skull base with a breach of the dura mater. Nontraumatic pneumocephalus is uncommon and can result from neurosurgical interventions, infection or tumors. In a minority of cases it can be due to barotrauma or frequent sneezing [1–4,6].

Very rarely, pneumocephalus can be secondary to a cerebrospinal fluid (CSF) fistula with leakage of CSF, classified as spontaneous pneumocephalus. A CSF fistula is an egress of CSF from the intracranial cavity through a defect or weak point in the skull. Most commonly they occur in the ethmoid roof, sphenoid sinus, and temporal bone. They can be

\textsuperscript{*} Author’s contributions: LVL: Conceptualization, Writing- Original draft preparation. SS: Supervision, Validation, Writing- Reviewing and Editing.

\textsuperscript{a} Corresponding author.

E-mail address: Said.Sanoussi@CHU-Brugmann.be (S. Sanoussi).
https://doi.org/10.1016/j.radcr.2020.07.036
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categorized into 3 groups according to their origin: congenital or acquired, further subdivided as nontraumatic (tumors or infections), traumatic (accidental or iatrogenic), or spontaneous [7–9]. Fistulas are categorized spontaneous when there is no predisposing or congenital etiology and thus, they are likely to be formed by bone remodeling or reabsorption during the patient’s life [8].

We report a case of spontaneous pneumocephalus associated with a CSF fistula at the right cribiform plate.

**Case report**

A 56-year-old woman presented to our emergency department with mainly bifrontal headache for 2-3 days which progressively increased in strength. The pain was described as an increase in pressure in the head. For 4 days she observed a clear liquid coming out of her nose.

The patient has a history of high blood pressure treated with olmesartan and medoxomil which she admits not to take on a daily base. She has no history of head trauma or previous surgery to the head and neck region.

At the emergency department, physical examination was normal. During neurological examination, there was no photophobia, no neck stiffness and Kernig’s sign was negative. She had no fever and laboratory findings were normal without signs for infection. A severely elevated blood pressure was measured (200/100 mm Hg) with normal pulse rate.

A noncontrast CT brain scan was performed to exclude an intracranial bleeding due to hypertension. The CT showed the presence of air (400-1000 UH) in the subarachnoid spaces, intraventricular and in the sellar region, predominant on the midline. Imaging also demonstrated an empty pituitary fossa. No intra- or extraparenchymal hemorrhage was detected. Mild inflammatory peripheral mucosal thickening in the maxillary sinuses and ethmoid cells was seen. No bone defect was visualized (Fig. 1). A β2-transferrin test was performed to confirm the presence of CSF in the nasal liquid.

The patient was hospitalized at the neurology department. A control CT scan performed the next day showed a resorption of most of the intracranial air. Magnetic Resonance, also performed the next day, didn’t show any relevant lesion. Therefore CT cisternography was performed to detect the exact location of the CSF fistula (6 mL of an iodinated non-ionic low-osmolar contrast agent injected in the thecal sac after lumbar puncture).

Cisternography showed the right gyrus rectus herniating inferiorly in the region of the right cribiform plate consistent with a small right-sided cribiform encephalocele. More anteriorly, a thin vertical opacified structure is demonstrated that joins the medial aspect of the ethmoid roof with the right cribiform plate consistent with contrast media leakage revealing a CSF fistula (Fig. 2).

The patient underwent neurosurgery with coverage of the defect. CSF leakage has been confirmed and rhinorrhea stopped completely after surgery.

**Discussion**

In a nontraumatic setting, the presence of pneumocephalus is very uncommon and often an incidental finding on CT imaging [2,4].

A CSF fistula can be suspected when careful history and physical examination have ruled out all other causes of nontraumatic pneumocephalus. The clinical diagnosis of a CSF leak is confirmed by obtaining a sample of nasal secretions and measuring the β2-transferrin activity [8–10]. The detection of β2-transferrin in nasal secretions was accurate in our case.

Since endoscopic surgery has become the standard treatment in small skull base defects, it is important to localize and characterize the bone defect and assess for possible encephalocele or meningoencephalocele [8,9,11].

Several imaging techniques have been used to diagnose and characterize skull base defects responsible for
CSF leakage. The main diagnostic imaging techniques include thin-section multidetector CT, MR cisternography, radionuclide cisternography and contrast-enhanced CT cisternography. Multidetector CT is often the first modality used to localize osseous defects [8,9,12].

In our case the CT findings could not demonstrate a skull base defect nor an air-fluid level or opacification of the contiguous sinus which are indicative of a CSF fistula [8,9].

Further evaluation with MR imaging did not demonstrate a meningocele or encephalocele, which are herniations of brain parenchyma or meninges through a bone defect [7–9].

In our case, further imaging with CT cisternography was performed as it is one of the standard methods for evaluating a possible CSF fistula [8,9,12]. CT cisternography could demonstrate the presence of a skull base defect and CSF fistula at the right cribiform plate.

Although CT cisternography is invasive, expensive, time-consuming with slight risk for complications, it proves its usefulness in cases with negative CT and MR results [8,9].

**Conclusion**

This case highlights the role of CT cisternography to identify and localize small osseous defects and CSF fistulas when CT and MR imaging findings are normal.

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