

Case Report



A Case of Non-Traumatic Pneumocephalus Associated with Otogenic *Proteus Mirabilis* Cerebellar Abscess

Mustafa CİHANGİROĞLU¹, İlhami ÇELİK², İsmail AKDEMİR³, Hakan ARTAŞ⁴, Ayhan AKBULUT²

¹Ergani Devlet Hastanesi, Enfeksiyon Hastalıkları ve Klinik Mikrobiyoloji Kliniği, DİYARBAKIR

²Fırat Üniversitesi Tıp Fakültesi Enfeksiyon Hastalıkları Anabilim Dalı,

³Harput Devlet Hastanesi, Beyin Cerrahi Kliniği,

⁴Elazığ Eğitim ve Araştırma Hastanesi, Radyoloji Kliniği, ELAZIĞ

ABSTRACT

In this case, it was presented a rare cerebellar abscess case due to *Proteus mirabilis* has an intraparenchymal gas formation at the early stage of cerebritis where gas formation disappeared at the late cerebritis phase of the abscess formation. ©2008, Fırat Üniversitesi, Tıp Fakültesi

Key words: Cerebellar apse, *Proteus mirabilis*, gas formation

ÖZET

Otojenik Kaynaklı *Proteus Mirabilis* İnfeksiyonu Sonucu Gelişen Serebellar Apse ile İlişkili Travmatik Olmayan Pnömocefalus Olgusu
Bu çalışmada, erken serebrit aşamasında parankim içi gaz oluşumu gözlenmesine karşın geç serebrit aşamasında bu görünümün kaybolduğu *Proteus mirabilis*'e bağlı nadir bir serebellar apse olgusu sunuldu. ©2008, Fırat Üniversitesi, Tıp Fakültesi

Anahtar kelimeler: Serebellar apse, *Proteus mirabilis*, gaz oluşumu

Brain abscess is a focal suppurative process within the brain parenchyma that continues to be a diagnostic and therapeutic challenge to clinicians (1). A brain abscess can result from direct extension of a cranial infection (e.g., osteomyelitis, mastoiditis, sinusitis, subdural empyema), penetrating head wounds, or hematogenous spread (e.g., in bacterial endocarditis, bronchiectasis, congenital heart disease with right-to-left shunt, or IV drug abuse (2). Both acute and chronic otitis media are considered as serious diseases in children because of their possible extracranial and/or intracranial extension (3).

Early presumptive clinical diagnosis supported by radiological evidence [computerized axial tomography (CAT) scan and magnetic resonance imaging] is the mainstay of diagnosis (4). *Proteus mirabilis* is usually implicated in neonatal and infantile meningitis, and brain abscess, especially in patients with otogenic infections (5). For years, it has been recognized that some brain abscesses are due to gas-forming organism and that plain radiographs may show an air-fluid interface. These infectious agents are usually *Clostridium* spp. and other gas-producing organisms with the mortality rate of about 25% (6).

In this case, we presented a gas-forming brain abscess of the early stage related with chronic mastoiditis where gas formation disappeared at the late cerebritis phase of the abscess

formation. *Proteus mirabilis* was cultured from the pus material provided at the time of mastoidectomy and observed its gas formation. This must be considered in differential diagnosis before starting the treatment in such cases with intraparenchymal pneumocephalus formed at the early stage of cerebral abscess.

CASE REPORT

Fifteen year-old-boy has been complained of intermittent fetid yellow discharge from the right ear canal past one year, he was brought in to the emergency department because of progressive headache and dizziness for the last four days. On initial physical exam, he was alert, cooperative and oriented, but acutely ill looking. At the time of the admission, temperature was found 36.5 °C, blood pressure at 100/60 mm/Hg, and heart rate at 64/min. On neurological examination, he had dysdiadokokinesia, dysmetria on finger to nose test, and signs of meningeal irritation. A fetid discharge mixed with debris was present in the right external ear canal. After the removal of the discharge, right eardrum was inspected. It was macerated and bulged outside. Physical and neurological examination findings were otherwise normal.

In laboratory parameters, hemoglobin was 11 g/dL, white blood cell count 11.000/mm³ (80% PNL, 14% lymphocytes, 6% monocytes), platelet count 263.000/mm³, erythrocyte sedimentation rate (ESR) 86 mm/h, CRP 46 mg/L, and ASO

1/400 IU/mL. On differential count, no toxic granulations were observed. Postero-anterior plain radiography of the chest x-ray was unremarkable.

Posterior fossa CT scans without contrast revealed a 2x5 mm lesion of air density located in the right cerebellar hemisphere and it was consistent with the pneumocephalus (Figure 1).

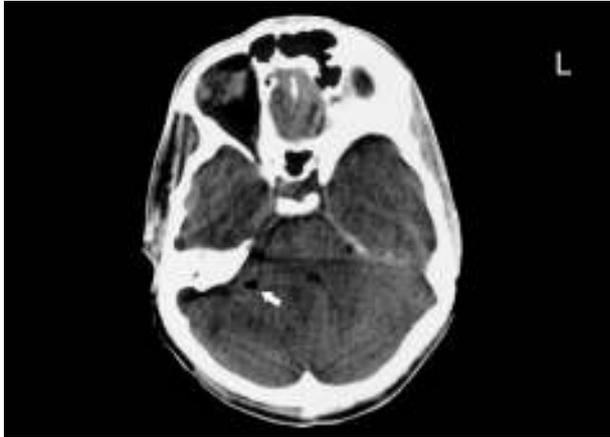


Figure 1. An air density localized on right cerebellar vermis (2x5 mm) (pneumocephalus)

A contrast CT a day later revealed an enhanced 8x4 mm ill-defined lesion compressing the fourth ventricle was demonstrated as well as pneumocephalus that was consistent with early cerebritis stage. An inflammation in mastoid was present which was filling out mastoid antrum destructing the wall and thereby extending into the posterior fossa. The air density (pneumocephalus) was observed adjacent to the mastoiditis lesion (Figure 2).

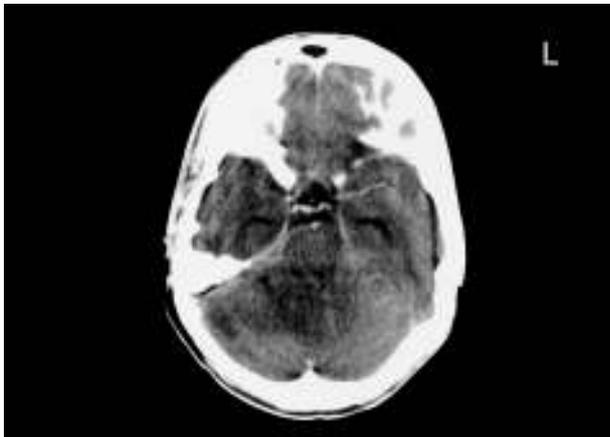


Figure 2. A compatible view of pneumocephalus and early cerebritis phase

Empirically, cefotaxime 3X2 g IV, ampicillin 4x3 g IV and metronidazole 4X500 mg/day IV therapy was implemented, and the patient underwent right radical mastoidectomy. Purulent material provided from the sigmoid sinus during the operation, which was analyzed bacteriologically, and proved the origin of *Proteus mirabilis* and any bacteria were obtained at anaerobic culture. Because the isolated microorganism was found to be sensitive to cefotaxime, ampicillin, therefore, cefotaxime was continued and ampicillin and metronidazole were discontinued.

Drowsiness, meningeal irritation signs, and bradycardia developed on the postoperative 5th day.

The follow-up contrast CT revealed, a biloculated mass lesion (daughter abscess) of 4x4 cm, which was thought to be consistent with cerebellar abscess, was found in the right cerebellar region extending to the tentorium and compressing the medulla posteriorly. Pneumocephalus, which was observed on the previous images, did not persist (Figure 3).

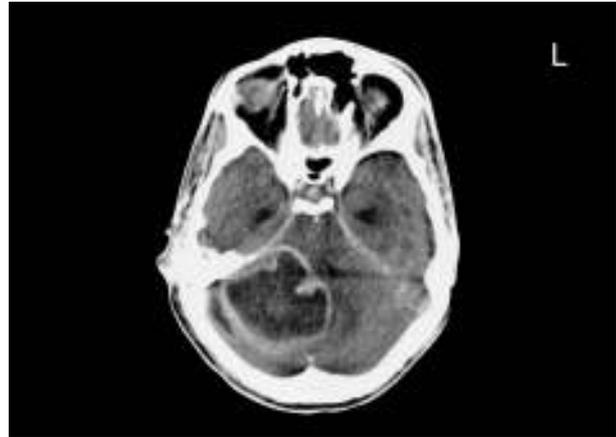


Figure 3. A compatible view of binoculated (daughter abscess) cerebellar abscess

An emergency aspiration was undertaken through a right occipital burr-hole under general anesthesia, and about 40 mL of thick yellow pus was evacuated. The patient developed neither postoperative complications nor fever. Contrast CT taken on 10th postoperative day demonstrated residual abscess formations in 4x3 and 2x1.5 cm diameters, which were extending from right pontocerebellar cistern to the occipital lobe. Fourth ventricle compression was still present. The patient was treated medically and observed clinically. On 33rd day of hospitalization, follow-up contrast CT displayed focal heterogeneous enhanced areas at the level of right cerebellar hemisphere, which were considered as residual inflammatory regions. The ventricular system and Sylvian fissures appeared normal (Figure 4).



Figure 4. Focal scattered contrasted fields at level of right cerebellar hemisphere (inflammation mixed with residue)

At the end of the 6-week therapy, the patient was clinically stable, and radiologically cleared from abscess formation. The white blood cell count was 7600/mm³, sedimentation rate 25 mm/h, and CRP 6 mg/L. Considering the

clinical improvement, the patient was discharged with a prescription of ampicillin 4x2 g P.O. and with cefuroxime axetil 250 mg 2x1 P.O. and instructed to come back at the end of 2 weeks of medical therapy.

At first month follow-up, no recurrence or residue was observed on CT scans, but hypodense postoperative changes at the site of previous abscess formation were observed (Figure 5). The physical and neurological findings of the patient were in better state at the first and 3rd day from the operation.



Figure 5. Postoperative hypodens changes at the localization of old abscess

DISCUSSION

Pneumocephalus is defined as entry of air into the cranial cavity, or intracranial gas collection. In a review of 295 cases of pneumocephalus, head injury and cranial surgery accounted for 74.5% of the cases (7). Infection by gas-forming organisms causes 8.8% of cases of pneumocephalus, the majority of which occur in patients with a predisposing factor such as trauma, sepsis, paranasal sepsis, and otitis media (8). Common causative organisms include *Clostridium perfringens*, *Escherichia coli*, *K. pneumoniae*, and *Enterobacter*, *Proteus*, *Pseudomonas*, *Peptostreptococcus*, and *Bacteroides* organisms (9).

The majority of brain abscesses originally stemmed from nasopharyngeal infections such as otitis media, mastoiditis, and sinusitis. Because of improved antibiotic therapy, today these sources account for approximately 40% of cases (10). Brain abscess occurring secondary to otitis media is usually localized to temporal lobe or cerebellum. In otogenic brain abscesses, the routine procedures are conservative or radical mastoidectomy, drainage of the abscess, incision of the sigmoid sinus and removal from its lumen of the clot, and broad-spectrum intravenous antibiotics therapy (3).

REFERENCES

- Whispelway B, Dacey RG, Scheld MW. Brain abscess. Scheld WM, Whitley RJ, Durack DT (eds). In: Scheld WM, Whitley RJ, Durack DT, eds. *Infections of the Central Nervous System*, 2nd ed. Philadelphia: Lippincott-Raven, 1997; 463-493.
- Tunkel AR, Whispelway B, Scheld MW. Brain abscess. In: Mandell GL, Douglas RG, Bennett JE (eds) *Principles and Practice of Infectious Diseases*, 5th edition. New York: Churchill Livingstone, 2000: 1016-1028.

Imaging in otogenic intracranial complications is considered as a diagnostic tool (11). The neuroimaging features of brain abscesses are vary with lesion stage. During the early cerebritis stage, NECT (Non-enhancement Computerized Tomography) scans may be normal or show only a poorly marginated subcortical hypodense area. In some cases, CECT (Contrast-enhancement Computerized Tomography) studies disclose an ill-defined contrast-enhancing area within the edematous region (12). On presentation of our case, CT displayed a 2x6 cm right cerebellar hemisphere lesion of air density, which was considered pneumocephalus, as well as the presence of ill-defined hypodense lesion consistent with the early cerebritis stage which was demonstrated on CT taken 2 days later, pneumocephalus location being entirely intraparenchymal, the absence of history of trauma, and isolation of *P. mirabilis* from the pus material from mastoidectomy region, all suggested the pneumocephalus to be of *P. mirabilis* origin.

Aerobic gram-negative bacilli are isolated in 23% to 33% of cases, usually in mixed culture, with *Proteus* spp. and *Pseudomonas aeruginosa* frequent isolates from abscess of otogenic origin (13). *Proteus* is included in the tribe Proteeae of the family enterobacteriaceae, Proteeae being ubiquitous in the environment and residing in the intestines of healthy humans and animals (14). Of the central nervous system infections, *P. mirabilis* is usually implicated in neonatal and infant meningitis, and brain abscess, especially in patients with otogenic infections (5).

Since confusion and intracranial pressure elevation were developed in the patient after 5 days of mastoidectomy, CT scans were provided and a 4x4 cm biloculated cerebellar mass (daughter abscess) being consistent with cerebellar abscess in the right cerebellar region extending to the tentorium and slightly compressing to the medulla was delineated. Pneumocephalus was not present which was observed on the previous CT scans. All reported pneumocephalus cases in the literature were with gas formation within the abscess cavity and at the stage of late capsulation, however, in this case, it was demonstrated in the early cerebritis stage (15-20). Besides, in this case there was no gas formation within the organized abscess. Prompt and proper antibiotherapy provided the abolishment of pneumocephalus, restricted the inflammatory process, and reduced the complication rate.

Pneumocephalus observed in patients with otogenic infection, should alert the clinician for the coexistence of a brain abscess that might have been caused by a gas-producing microorganism; and therapy should be implemented promptly.

- Kuczowski J, Mikazewski B. Intracranial complications of acute and chronic mastoiditis: report of two cases in children. *Int Pediatr Otorhinolaryngol* 2001; 60: 227-237.
- Mathisen GE, Johnson JP. Brain abscess. *Clin Infect Dis* 1997; 25: 763-781.
- Renier D, Flandin C, Hirsch E, et al. Brain abscess in neonates: a study of 30 cases. *J Neurosurg* 1988; 69: 877-882
- Candan S, Katelioglu M, Ceylan S, Koksali I. Otogenic brain abscess with pneumocephalus. *Infection* 1990; 18: 191-192.

7. Andrews JC, Canalis RF. Otogenic pneumocephalus. Laryngoscope 1986; 96: 521-528.
8. Frankel M, Faley D, Alker G: Otogenic pneumocephalus secondary to chronic otitis media. Arch Otolaryngol 1980; 196: 437-439.
9. Liliang PC, Hung KS, Cheng CH, Chen HJ, Ohta I, Lui CC. Rapid gas-forming brain abscess due to Klebsiella pneumoniae. Case illustration: J Neurosurg 1999; 91: 1060
10. Alderson D, Strong AJ, Ingham HR, Selkon JB: Fifteen-year review of the mortality of brain abscess. Neurosurgery 1981; 8: 1-6
11. Lunz M, Keren G, Nussem S, Kronenberg J. Acute mastoiditis-revisited. ENT J 1994; 73: 648-654.
12. Osborn GA. Infections of the brain abscess and its lining. Osborn GA (ed.). IN: Diagnostic Neuroradiology, Mosby Inc. Missouri, USA. 1994: 673-715.
13. Heilpern KL, Lorber B. Focal intracranial infections. Infect Dis Clin North Am 1996; 10: 879-898.
14. Aleksic S, Bockem Ühl J. Yersinia and other Enterbacteriaceae. In: Murray PR, Baron EJ, Pfaller MA, et al.(eds.) Manual of Clinical Microbiology. Washington DC: ASM Press, 1999: 483-496
15. Norrell H, Howieson J. Gas-containing brain abscesses. Am J Roentgenol Radium Ther Nucl Med 1970; 109: 273-276
16. Davini V, Rivano C, Tercero E. Spontaneous pneumocephalus caused by otogenous temporal abscess. J Neurosurg Sci 1974; 18: 210-215
17. Azar-Kia B, Sarwar M, Batnitzky S, Schechter M. Radiology of intracranial gas. Am J Roentgenol Radium Ther Nucl Med 1975; 124: 315-323
18. Taguchi Y, Sato J, Nakamura N. Gas-containing brain abscess due to Fusobacterium nucleatum. Surg Neurol 1981; 16: 408-410.
19. Young RF, Frazee J. Gas within intracranial abscess cavities: an indication for surgical excision. Ann Neurol 1984; 16: 35-39.
20. Vaquero J, Martinez R. Tension pneumocephalus in brain abscess after simple puncture-drainage. Acta Neurochir 1984; 71: 225-227

Kabul Tarihi: 29.05.2007