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Case Report

Otogenic Sinus Thrombosis with Air Embolism - A Case Report

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Abstract

Cerebral venous air embolism (CVAE) is an extremely rare clinical entity, usually associated with certain iatrogenic procedures. To the best of our knowledge there are no reported cases of ear-related cerebral venous air embolism in the up-to-date literature. However, although quite uncommon in the antibiotic era, cerebral venous sinus thrombosis is a well known intracranial complication of otitis media. We are presenting a clinical case of a 53-year-old male patient who was treated at our department and was diagnosed with both of these rare conditions. Possible mechanisms that could contribute to the clinical presentation will be discussed and literature briskly reviewed. As the infections in the head and neck region account for only a minority of all cerebral venous sinus thrombosis (CVST) cases, other risk factors must be taken into consideration.

Keywords: Sinus Thrombosis; Cranial Sinus Thrombosis; Middle Ear Effusion; Air Embolism; Embolism, Gas

Introduction

Otogenic CVST is an uncommon complication of the middle ear disease and results from either the direct spread of infection through a dehiscent temporal bone or thrombophlebitis of mastoid emissary veins draining to the sigmoid sinus. It can be a consequence of the chronic otitis media with or without cholesteatoma or an aggressive form of the acute otitis media (AOM), the latter nowadays being the case mainly in paediatric patients [1,2]. Signs and symptoms of the CVST depend on the severity of the disease. In most cases, patients present with a fever, headache, otalgia, otorrhea, vomiting and pain in the neck. Luminal occlusion causing disruption in the cerebral venous drainage can result in a raised intracranial pressure presenting as papilledema. Griesinger sign is pathognomonic for the sigmoid sinus thrombophlebitis and is described as tenderness and swelling over the mastoid plane. Clinical features can be masked by prior antibiotic treatment, therefore the patient may be relatively unaffected. Throughout history, the incidence of sigmoid sinus thrombosis has changed from being the second most common intracranial complication (IC) in the pre-antibiotic era to being a rare finding nowadays, when it accounts for around 6% of all IC otogenic complications [3]. According to the literature, head and neck infections are responsible for only 8% of all CVST, while 12,5% of them are idiopathic. Other risk factors include oral contraceptives, thrombophilia, malignancy and haematological diseases [4].

Cerebral venous air embolism is observed less frequently than cerebral arterial air embolism and is associated with high morbidity and mortality. Pathophysiology is still not completely clear. Most often it is associated with certain iatrogenic procedures: central or peripheral vein catheter placement, surgery, endoscopy and haemodialysis. Physical preconditions for the direct entry of gas into the venous system are mural impairment of non-collapsing veins and the presence of the subatmospheric pressure in these vessels. Non-collapsing veins include the epiploic and emissary veins plus the dural venous sinuses [5]. According to the literature, an injection of 2 to 3 mL of air into the cerebral circulation can cause neu-

rologic deficits such as altered mental status, seizures, coma, transient focal neurological deficits and may be fatal [6,7]. It is difficult to estimate the true incidence as the course is often subclinical or the symptoms are non-specific. Besserau., *et al*. estimated the incidence of the iatrogenic air embolism, both arterial and venous, at 2.65 per 100 000 hospital admissions [8].

Case Description

A 53-year old male patient presented to the emergency neurologic clinic after a series of three partial epileptic seizures lasting 1-3 minutes and a left-sided headache. His medical history revealed type-2 diabetes with diabetic neuropathy and arterial hypertension. Parenteral levetiracetam was applied, stopping the seizures altogether. A non-contrast CT head revealed significant air embolism in the sagittal and left sigmoid sinus. There was also fluid in the middle ear and some of the mastoid cells on the same side, but no signs of mastoiditis or AOM were observed (Figure 1). The inflammatory markers were mildly elevated (CRP 67, WBC 11,7). He was referred to our ENT department where an additional anamnesis revealed that he had been receiving oral amoxicillin-clavulanic acid (AMX-CLA) in the week prior to the admission, which had been prescribed for left-sided otalgia by his family physician. Otoscopy revealed signs of serous middle-ear effusion on the left side. Audiometry showed a left-sided combined hearing loss with an air-bone gap of 20-40 dB. Intravenous AMX-CLA was reintroduced. The following day, a contrast-enhanced head MRI and MR venography was performed, which revealed a massive venous thrombosis of the sagittal, both transverse and sigmoid sinuses accompanied by extensive air bubbles (Figure 2). Middle-ear and mastoid effusion was noted on both sides, although it was more apparent on the left. A neck doppler ultrasound excluded the propagation of the thrombus into the jugular veins. He began receiving therapeutic doses of low-molecular-weight heparin (LMWH) and the antibiotic was switched to cefotaxime. We employed conservative measures (oxygen, bed rest, negative Trendelenburg position). After one week, a follow-up CT imaging showed air resorption with a mild spread of thrombus noted intracranially. The patient required an increased dose of levetiracetam due to some minor seizure activity. A left tympanostomy was performed and serous fluid was drained from the ear. Microbiological analysis showed that the sample was sterile. No further symptoms were noticed. He was discharged on the 14th day with LMWH and antiepileptics.

Figure 1: Head CT revealing significant air embolism in the sagittal and left sigmoid sinus (yellow arrows). Fluid is present in the middle ear and some of the mastoid cells on the same side (red arrow), but no signs of mastoiditis.

Figure 2: Contrast-enhanced head MRI and MR venography revealed a massive venous thrombosis of the sagittal, both transverse and sigmoid sinuses accompanied by extensive air bubbles (blue arrows). Middle-ear and mastoid effusion on both sides, more apparent on the left (red arrows).

A follow-up contrast-enhanced head MRI and MR venography were performed 3 months later. The study revealed partial recanalisation of the sagittal, both transverse and sigmoid sinuses with

some residual filling defects. No cerebral or cerebellar ischemia and oedema were noted. There was still a noticeable effusion and mucosal thickening of mastoid cells on both sides, yet still more pronounced on the left (Figure 3). During the follow-up visit, the patient complained about occipital headaches and occasional speech difficulties. Otomicroscopy showed that the tympanostomy tube was patent and the ear was dry. We consulted a neurologist, who suggested continuing with the anticonvulsant and anticoagulant therapy for a total duration of six months.

Figure 3: MRI and MR venography performed 3 months later. Partial recanalisation of the sagittal sinus with some residual filling defects (blue arrow). Noticeable effusion and mucosal thickening of mastoid cells on both sides, yet still more pronounced on the left (red arrow).

Discussion

The following aspects of the presented case should be discussed: the source of air embolism, the etiology of thrombosis and the cause of persistent mastoid effusion that was noted on the follow-up MRI scan. When the patient was treated at the neurology department right after the seizures, a peripheral vein catheter was placed, which might have caused the air embolism. Considering a possibility that the thrombosis was not noted on a non-contrast CT scan (NCCT), this could be the case of a coincidental iatrogenic air embolism and otogenic CVST. A study from 2009 showed that a non-contrast CT scan detection sensitivity and specificity for cerebral venous thrombosis are 64.4% and 97.2% respectively [9].

Schlimp., *et al*. explored the movement of air bubbles from peripheral veins to the cerebral sinuses and described the retrograde venous pathway in an upright positioned patient. Some anatomical traits such as patent foramen ovale or presence of pulmonary shunt can further increase the risk of this mechanism occurring [10-12]. The other theoretically possible route of air entry into the sigmoid sinus is through the dehiscent adjacent mastoid bone, though we were not able to find any published case depicting a direct passage of atmospheric air into the venous lumen in a patient with a middle ear infection. In order for luminal air entry to happen there would have to be a direct communication between the atmospheric air and non-collapsing venous lumen, as well as an adequate pressure gradient of at least 5 cm H_2O [5,6]. We also found two case reports of the Lemierre syndrome, where the authors described air bubbles found on CT scan and attributed the condition to gas-producing anaerobic bacteria [13,14]. Considering the patient's significant amount of air inside the venous sinuses and negative middle ear swab culture, we believe that this option is not very likely.

In our opinion, the most probable cause of CVST is AOM, as we could not identify any other risk factors. By the time of our inspection, the clinical picture was probably masked as the patient had already received antibiotics prior to admission. The persistent bilateral mastoid effusion was most likely the result of a venous congestion due to the posterior fossa CVST [15]. Only partial recanalisation of venous sinuses had occurred until the follow-up exam as it was already described.

Conclusion

As a coincidence of three separate clinical entities is very unlikely (AOM, CVAE, CVST), the concept of Occam's razor points to the AOM as the likely culprit, with the CVAE being iatrogenic. We cannot fully exclude that the CVST of an unknown origin was the initial event, with the middle-ear effusion and headache being mistaken for the AOM.

We hope that our case report could serve as an aid in the thinking process when dealing with these rare entities.

Bibliography

1. Harker LA and Clough S. "Complications of temporal bone infections". In: Cummings C, ed. Otolaryngology Head and Neck Surgery. 4th ed. Philadelphia, PA: Mosby, Inc. (2005).

2. Johnston DR., *et al*. "Sigmoid sinus thrombosis as a complication of otitis media". *Otology and Neurotology* 28.7 (2007): 988-989.

3. Viswanatha B and Naseeruddin K. "Lateral sinus thrombosis in otology: a review". *Mediterranean Journal of Hematology and Infectious Diseases* 2.3 (2010): e2010027.

4. Ferro JM., *et al*. "Prognosis of cerebral vein and dural sinus thrombosis: results of the International Study on Cerebral Vein and Dural Sinus Thrombosis (ISCVT)". *Stroke* 35.3 (2004): 664-670.

5. Muth CM and Shank ES. "Gas embolism". *The New England Journal of Medicine* 342.7 (2000): 476-482.

6. Ho AM. "Is emergency thoracotomy always the most appropriate immediate intervention for systemic air embolism after lung trauma?" *Chest* 116.1 (1999): 234-237.

7. Giraldo M., *et al*. "Venous air embolism in neurosurgery". *Colombia Journal of Anesthesiology* 43 (2015): 40-44.

8. Bessereau J., *et al*. "Long-term outcome of iatrogenic gas embolism". *Intensive Care Medicine* 36.7 (2010): 1180-1187.

9. Linn J., *et al*. "Noncontrast CT in deep cerebral venous thrombosis and sinus thrombosis: Comparison of its diagnostic value for both entities". *AJNR American Journal of Neuroradiology* 30.4 (2009): 728-735.

10. Costa Carneiro A., *et al*. "Cerebral Venous Air Embolism: A Rare Phenomenon". *European Journal of Case Reports in Internal Medicine* 6.61 (2019): 1.

11. Schlimp CJ., *et al*. "Cerebral venous air embolism: what is it and do we know how to deal with it properly?" *JAMA Neurology* 71.2 (2014): 243.

12. Schlimp CJ., *et al*. "The potential of venous air embolism ascending retrograde to the brain". *Journal of Forensic Science* 50.4 (2005): 906-909.

13. Turan A., *et al*. "A case of lemierre syndrome secondary to otitis media and mastoiditis". *Case Reports in Emergency Medicine* 2014 (2014): 208960.

14. Riordan T. "Human infection with Fusobacterium necrophorum (Necrobacillosis), with a focus on Lemierre's syndrome". *Clinical Microbiology Review* 20.4 (2007): 622-659:00011-07.

15. Shah LM., *et al*. "Mastoid findings secondary to posterior fossa dural venous sinus thrombosis". *American Journal of Roentgenology* 201.2 (2013): 406-411.