CASE REPORT

Arterial brain infarction in complicated acute otitis media

Yael Oestreicher-Kedem,*, Liat Ben-Sira, Galia Grissaru, Ari DeRowe

Pediatric Otolaryngology Unit, Dana Children's Hospital, Tel-Aviv Sourasky Medical Center, Sackler Faculty of Medicine, Tel-Aviv University, Tel-Aviv, Israel

Department of Radiology, Tel-Aviv Sourasky Medical Center, Sackler Faculty of Medicine, Tel-Aviv University, Tel-Aviv, Israel

Infectious Disease Unit, Dana Children's Hospital, Tel-Aviv Sourasky Medical Center, Sackler Faculty of Medicine, Tel-Aviv University, Tel-Aviv, Israel

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Summary
We report a rare case of acute otitis media (AOM) complicated by arterial brain infarction, meningitis and orbital cellulitis. Computed tomography (CT) initially showed suspected epidural abscess, coalescent mastoiditis, and orbital cellulitis. Further clinical deterioration occurred following mastoidectomy and evacuation of the abscess. Magnetic resonance imaging (MRI) revealed subacute brain infarction and an extra-axial brainstem abscess. Revision of the initial CT revealed subtle signs suggestive of arterial brain infarction. Although CT with contrast is the standard of care for suspected AOM complications, MRI should be considered as an adjunct when CT is not definitive and neurological signs are present.

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1. Background

We describe a case of acute otitis media (AOM) with multiple complications, notably arterial brain infarction. About 2% of cases of AOM will develop intracranial complications, most commonly meningitis [1,2]. Focal or widespread brain infarctions, as sequela of bacterial meningitis, are usually venous, and have been demonstrated in 8–27% of children with bacterial meningitis [3–5]. Arterial brain infarctions are, however, rarely mentioned as a direct complication of AOM.

2. Case presentation

A 30-month old girl was brought to the pediatric emergency department due to fever, earache and right ear discharge of 2 weeks duration. Her
pediatrician had diagnosed AOM and put her on a 7-day course of amoxicillin-clavulanic acid per-os (p.o.) and ciprofloxacin eardrops. The fever and ear discharge subsided, but reappeared 3 days later. She had no significant past medical history and had received all age-appropriate vaccinations. Her vital signs at presentation were temperature 38.2 °C, heart rate 156/min, and oxygen saturation 99% in room air. Physical examination revealed an alert but pale child with a right ear purulent discharge and erythematous pharynx. There was no redness or swelling in the post-auricular region. The neurological examination was normal. A central perforation of the tympanic membrane was revealed after the pus from the ear canal had been suctioned. Cultures were obtained of the draining pus and blood. Hemoglobin level was 9.3, white blood cell count 17,300, neutrophils 88.4%, C-reactive protein 270 and sedimentation rate 100 mm/h. She was admitted to the pediatric department and was started on intravenous (i.v.) ceftriaxone and ciprofloxacin eardrops.

Her condition did not improve, and swelling of the right eyelids and mild ptosis, diagnosed as preseptal cellulitis, appeared the following day. Later that day, she started vomiting and became stuporotic. Neurological examination revealed nuchal rigidity and reduced tonus, while the fundus examination, cranial nerves and Babinski signs were normal. A non-contrast computerized tomographic (CT) scan of the head was performed and showed lack of aeration of the right mastoid cavity and soft tissue swelling of the mastoid and middle and external ear. The brain CT scan at that time was interpreted as normal, with no midline shift, bleeding or focal findings. A lumbar puncture yielded CSF with 10,800 white blood cells (96% neutrophils), 20 red blood cells, glucose level 2 mg/dl and protein level 127 mg/dl. CSF gram stain showed no bacteria. The child was diagnosed as having bacterial meningitis and vancomycin i.v. was added to the antibiotic treatment. CSF and blood cultures showed no growth. The discharge from the ear was cultured and grew *Diphtheroids*.

On the third day, there was still no improvement in her fever and ear drainage, and the swelling of the right eye worsened. A contrast CT scan of the head showed soft tissue swelling of the right mastoid cavity, middle and external ear, bone lysis of the mastoid wall adjacent to the sigmoid sinus and a suspected small epidural abscess lateral to the sigmoid sinus (Fig. 1). There was no visible filling defect adjacent to the sigmoid sinus. Severe right peri orbital swelling was noted, with retrobulbar infiltration, enlargement of the recti muscles (predominantly the superior rectos), and dilatation of the right ophthalmic vein. The brain scan was interpreted as normal. She was taken to the operating room and underwent a cortical mastoidectomy, evacuation of a perisinus epidural abscess and ventilation tube insertion. Puncture and aspiration of the sigmoid sinus revealed blood. Metronidazole i.v. was added to the antibiotic therapy. One day postoperatively, her fever went down to 38 °C and the eye swelling improved.

Right gaze preference and hyperreflexia appeared during the next few days. A repeated lumbar puncture revealed CSF with 388 white blood cells (43% neutrophils), glucose level 30 mg/dl and protein level 146 mg/dl. A magnetic resonance imaging (MRI) scan of the head was performed on the seventh day after admission and revealed a subacute brain infarct in the territory of the right posterior cerebral artery (RPCA), with subdural abscesses along the cisterns of the right cerebropontine angle, quadrigemina and ambien, compressing and obliterating the RPCA (Fig. 2). Enhancement of the non-aerated right mastoid, fatty infiltration of the orbit, and enhancement of the right eighth and ninth cranial nerves up to the internal auditory canal were noted. A magnetic resonance angiogram (MRA) showed only the proximal part of the RPCA (Fig. 3) with acute cutoff. Following the MR findings, the initial head CT scan was reviewed and, in retrospect, similar changes were observed in the brain parenchyma in the distribution of the RPCA.

Sub-coetaneous enoxaparin was added to the drug regimen and the combination of antibiotics was continued. A second MRI, performed 2 weeks...
later, demonstrated an old brain infarction in the territory of the RPCA involving the right occipital lobe, thalamus and midbrain. The subdural abscesses were now smaller. The MRV was normal.

The patient’s condition gradually improved, the ear discharge stopped with the ventilation tube in place, and her temperature normalized on hospital day 12. Altogether, she had received 6 weeks of i.v. antibiotic treatment followed by 6 weeks of amoxicillin-clavulanic acid p.o. A repeat neurological examination before discharge showed normal motor strength, bilateral symmetric hyperreflexia, and mild right eye ptosis. The rest of the neurologic exam was normal. Pure tone audiometry and speech reception thresholds were within normal range. Immunoglobulin levels were normal. The hypercoagulability workup had initially revealed low protein S level (31%) that became normal 3 months later (64%), suggesting that the patient had no prothrombotic tendency. Enoxaparin was stopped and she received only low-dose aspirin. An MRI performed 3 months after discharge showed resolution of the brain infarction in complicated acute otitis media.
subdural abscesses with an old infarct in the territory of the RPCA (Fig. 4).

3. Discussion

This report demonstrates a case of AOM with multiple intracranial sequelae, consisting of meningitis, arterial brain infarction, brain abscess, orbital cellulites, and most notably, arterial brain infarction, a rare complication.

Brain infarctions occur much less frequently in children than in adults, but the role of infectious and inflammatory causes of stroke is much more significant in the pediatric population [6]. Atherosclerosis, a major underlying disorder of stroke in adults, is absent in children [7]. Non-infectious risk factors for arterial infarction in children are cerebral arterial abnormalities, previous v. zoster infection, preceding trauma and anemia, cardiac abnormalities and thrombophilia [8]. The risk factors are often multiple and age related [7].

Two major types of vascular injuries may lead to brain ischemia and infarction: arterial ischemia resulting from occlusion within the arterial supply to the brain, and sinovenous thrombosis related to occlusion in the venous drainage of the brain [7]. Sinovenous thrombosis is a more typical complication of otitis media, mastoiditis and meningitis than arterial ischemia [9]. In our case, however, as demonstrated by MRA, the brain infarction was caused by RPCA occlusion. Several of the mechanisms of stroke development in children described by Takeoka and Takashi [6] could have contributed to the development of ischemic arterial brain infarction in our patient with otitic meningitis: (1) spread of the meningeal inflammation to involve the walls of the intracranial vessels, resulting in arterial thrombosis with ischemia; (2) direct compression on the RPCA by the subdural fluid collection; (3) septic shock resulting in systemic hypotension contributing to the cerebral ischemia (usually diffuse brain ischemia); (4) a hypercoagulable state due to an inherited prothrombotic tendency. Although we first suspected a prothrombotic tendency, due to her initial low protein S level during the acute stage, it normalized later on. The proximity of the subdural collection to the RPCA makes it more likely for compression of the RPCA to have been the etiology for the subsequent arterial thrombosis and occlusion.

Periorbital cellulitis is another infrequent complication of AOM which was present in our patient. Otitis had been reported as a predisposing factor for periorbital cellulites [10,11] either by contiguous spread from the ear (probably through zygomatic air cells) or by systemic bacteremia [10]. In children younger than 5 years, as in our case, periorbital cellulitis is more often associated with upper respiratory tract infection and positive blood cultures [11]. Meningitis can also occur as a complication of bacterial periorbital cellulitis [12].

Early diagnosis and treatment of complications of otitis and meningitis are essential for improving outcome. CT is often used in the early workup of meningitis to rule out intracranial pressure before carrying out lumbar puncture, for ruling out complications of meningitis and in cases of acute mastoiditis unresponsive to treatment. CT is often the first imaging study used due to its availability and cost. It can demonstrate major complications of meningitis, such as brain edema, late infarcts, hydrocephalus, subdural effusions and brain abscesses [13]. As demonstrated in our case, however, it may be insensitive to the subtle and early parenchymal ischemic changes that appear in early brain infarcts. Therefore, other imaging modalities should be considered when there are neurological signs that cannot be explained by the findings in the brain CT or in cases non-responsive to treatment. MRI is more sensitive than CT in diagnosing inflammatory vasculitis and the extent of parenchymal damage, especially in early infarcts that can be demonstrated with diffusion-weighted imaging (DWI). By delineating the cytotoxic edema (bright signal) in the early stage of acute ischemia or infarction, DWI can detect acute infarction within minutes of onset [13]. It also has the ability to distinguish between extra-axial empyemas (high signal on T1) versus reactive effusions (low signal on T1) [13]. MRA can supplement MRI for demonstrating arterial flow, without exposure to the additional radiation of CT. Conventional angiography should be considered to rule out an intracranial vascular abnormality (present in up to 50% of children with arterial ischemic stroke) when a non-infectious condition for brain infarction is suspected [7].

Empiric antibiotic treatment of bacterial meningitis usually includes a third-generation cephalosporin plus vancomycin [14], with further modification according to cultures and sensitivity. Brain abscess secondary to otic infection usually involves mixed flora [15], in which case a third-generation cephalosporin and metronidazole is recommended. In our case, ceftriaxone was administered first, and vancomycin was added to cover cephalosporin-resistant Streptococcus pneumoniae after bacterial meningitis had been diagnosed. Metronidazole was added to cover anaerobes when she was found to have a brain abscess. We could not isolate the causative organism, probably due to prior antibiotic treatment. The benefit of adding
dexamethasone to the treatment in children with bacterial meningitis in order to improve neurological outcome has not yet been determined [14,16], and we did not administer it.

Treatment should also address the source of infection (e.g., otitis) and complications of meningitis (e.g., brain abscess and infarction). In our case, treatment included cortical mastoidectomy, evacuation of a perisinus epidural abscess and ventilation tube insertion. It was decided not to drain the subdural abscess, but rather to wait for response to the antibiotic treatment with close imaging follow-up. This approach to brain abscesses is usually kept for selected cases in which the duration of illness is less than 2 weeks, when the abscess diameter is less than 2 cm and when there are no neurological deficits and no signs of increased intracranial pressure [15,17]. Subsequent MRI scans demonstrated resolution of the abscesses in our patient. We added enoxaparin to the treatment to lower the risk of recurrent arterial brain infarction, and because of suspicion of a prothrombotic tendency (protein S deficiency) [18]. When the child’s protein S levels normalized, the regimen was switched to long-term aspirin to prevent recurrence.

4. Conclusions

Despite the availability of antibiotic treatment, AOM can still lead to life-threatening and debilitating complication, such as arterial brain infarction. Neurological signs and lack of clinical improvement under appropriate antibiotic and surgical treatments should raise the suspicion of intracranial complications. CT scans of the brain, especially non-contrast ones, are not sufficient to rule out early neurological complications, as we demonstrated herein. Thus, DWI and MRI should be considered for early detection of these intracranial complications.

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References