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Adult Primary Ventriculitis as a complication of acute otitis media: A comprehensive review of reported cases

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ABSTRACT

Introduction: Acute primary bacterial ventriculitis in adults is a rare intracranial disease. It can be a complication of neurosurgical patients with ventricular stent insertions or in children.
Objective: This paper presents a case of acute otitis media in a 71-year-old diabetic male that progressed rapidly to acute ventriculitis, with a literature review of presentation, work up, management and patient outcomes.
Methods: A search using MEDLINE and EMBASE was carried out including "primary ventriculitis", "bacterial ventriculitis" or "pyogenic ventriculitis" in the adult population. The cases were summarised.
Results: A total of 13 case reports were analysed. There was only one other case of pyogenic ventriculitis presenting with of sudden onset hearing loss, which turned out to be a complication of ventriculitis. Common presenting symptoms included agitation, depressed consciousness but no case reported any signs of meningism. This is the only known case of ventriculitis following acute otitis media.
Conclusion: Ventriculitis can result as a complication of otological disease; it can manifest as rapid neurological deterioration and is difficult to diagnose. A high index of suspicion should be held for ventriculitis in cases with rapid progression or severity. Optimal work up includes serial MRI and lumbar puncture, for prolonged, targeted antimicrobial therapy.

1. Introduction

Ventriculitis is a known complication of ventricular shunt insertion, intracranial pathology or trauma and can carry a high mortality. Ventriculitis following neurosurgical procedure can be as high as 45% [1]. Acute bacterial ventriculitis unrelated to trauma or neurosurgery is a rare occurrence in adults, though it does occur in immunocompromised children. At time of writing, there are less than 15 such reported cases of ventriculitis, the first recorded in 1977. There are varying presentations such as fever, agitation or depression of consciousness and of headaches making early diagnosis challenging [1,2]. Causative microbes vary greatly due to a range of aetiologies. Otogenic meningitis is well documented with the prevailing causative organism being *Streptococcus pneumoniae* [3].

Magnetic Resonance Imaging (MRI) features of ventriculitis include ventricular debris, hydrocephalus and hyperintense periventricular signals, and ependymal enhancement. It has been suggested that in cases of a meningitis-type picture without response to initial therapy, a diagnosis of pyogenic ventriculitis should be considered and confirmed with cerebrospinal fluid (CSF) culture and MRI [1–5].

We present a unique case of pyogenic ventriculitis that arose from acute otitis media and a review of literature.

2. Methods

A comprehensive literature review was conducted using Embase and Medline databases. The primary objective was to identify documented cases of ventriculitis secondary to a bacterial insult, where possible the antimicrobial therapy received and the patient outcome. 13 cases of secondary ventriculitis were identified and summarised.

3. Case report

A 71-year-old male presented to the emergency department with worsening headache and agitation. He had been unwell and complaining of a painful sensation in the ear that then "popped". He had visited the GP earlier in the day who diagnosed and commenced treatment for acute otitis media. He had a background of hypertension and

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hypercholesterolemia. The patient was intubated for fluctuating Glasgow Coma Scale (GCS). Initial Computed Tomography (CT) imaging showed evidence of intracranial infection with gas forming organism. CT with contrast showed meningitis as a complication of otomastoiditis and a defect in the right tegmen tympani. The patient remained sedated and was admitted to Intensive Care Unit (ICU). Further investigations of lumbar puncture and MRI followed. The patient had a purulent lumbar puncture, with normal opening pressures. The CSF cultures grew streptococcus pneumoniae. MRI revealed extensive Fluid-attenuated inversion recovery (FLAIR) signal of the leptomeninges and in the sulci, most marked on the right frontal sulci in keeping with meningitis. There was also evidence of layering debris in the occipital horns of the lateral ventricles and diffusion restriction consistent with ventriculitis (see Figs. 1.1 and 1.2). The day following admission the patient had a right myringotomy and grommet inserted, pus discharge that also grew Streptococcus pneumoniae.

Extubation was delayed by ongoing seizures. The patient required dual anti-epileptic therapy of phenytoin and Levitiracetam and treated with 4 days of intravenous glucocorticoids for seizure control. A repeat CT head showed evidence of cerebritis or micro-infarcts that had developed 5 days after the initial CT head.

On extubation, he had reduced movement of the left arm, expressive dysphasia, and global weakness probably due to ongoing infective process, reflected in low-density changes on serial CT. He was treated with intravenous Meropenem as per CSF culture sensitivities and began physiotherapy rehabilitation. On discharge he was making improvement with his speech and left arm, but still globally weak. He was discharged from ICU to complete a 6-week course of intravenous meropenem, and on Levitiracitam for seizure control.

4. Results

There were 14 cases that described the work up and management of patients with primary ventriculitis. The management of cases are summarised below in order of patient age, with our case in bold.

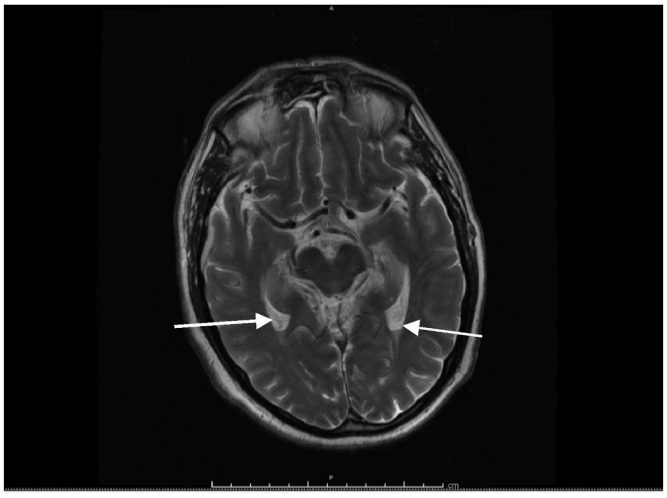

Fig. 1.1. MRI showing layering debris (arrows) within the occipital horns of the lateral ventricles with diffusion restriction consistent with ventriculitis [1].

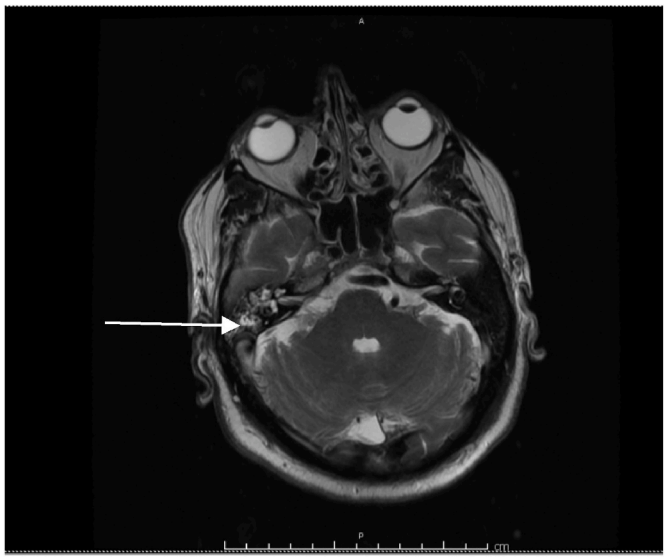

Fig. 1.2. There is also evidence of diffusion restriction within the right mastoid/middle ear and left maxillary antrum in keeping with active infection (arrow).

5. Discussion

The ages of patients described range from ages of 39–85 with 3 mortalities, aged 62–81. 3 of the 14 patients were female. The diagnosis of ventriculitis appears difficult due to non-specific signs and varied patient characteristics. Including our case, common themes reported included: fever (71%) with agitation of depression of consciousness (28%), headache (28%), issues with hearing (21%), and focal neurology such as hemiplegia or seizures (14%). A marked and rapid progression of disease was reported in all cases. There was an absence of meningism in 12 out of 13 of cases reported.

The radiological diagnosis of ventriculitis is the unifying and reliable theme of the more recent case reports reviewed. A case reported in 1977 where the patient was initially admitted with hemiplegia and the imaging of choice was Tc-pertechnetate, which infers that improved MRI accessibility since that time has increased diagnosis of this disease [10]. Fukui et al. showed that layering and debris within the ventricles was specific, with other non-specific signs of hydrocephalus and periventricular high signalling.

The case described by Ito H et al. showed all three signs of intraventricular hyperdensity, hydrocephalus and periventricular signal. The patient was surgically managed, with external ventricular drain, but later died. Ventricular shunt as successful treatment was described in two other cases by Vajramani et al. and Barloon et al. Due to the low case load of patients there are no best practice guidelines as to when to consider ventricular shunt. Our patient was discussed with neurosurgery and was deemed unsuitable for surgical intervention, but to revisit options if the patient deteriorated or developed hydrocephalus.

It appears the most important aspects of treating primary ventriculitis is to obtain a CSF organism to begin targeted antibiotics early. Case reports without mortality gave a targeted therapy early and for average duration of 30 days, ranging from survival and improvement after 14 days up to 49 days. Optimal treatment duration remains unclear but can be guided by clinical picture, serial bloods and lumbar puncture, or serial imaging [6,7].

Age & gender	Presentation	CSF culture	Treatment (days) and outcome	Author
85 Male	Fall at home, fever and seizure in ED	*Neisseria meningitidis*	IV cefotaxime [14] Followed by oral lefloxacin (24) Dexamethasone Survived	Louasard A et al. 2018 [2]
81 Female	Right hemiplegia	*Staphlococcus aureus*	Oxacillin [3] Chloramphenicol [3] Died	Kyung Lee H 1977 [9]
73 Male	Disturbance of consciousness, fever and headache	*Streptoccocus viridans*	Died, ventriculitis diagnosed on autopsy	Yamamoto F 2017 [19]
71 Male	Dysuria, nausea, fever, fluctuating consciousness	*Escheria coli*	Ceftriaxone (42)	Lopes JC et al. 2020 [16]
71 Male	**Fever, agitation following otitis media**	***Streptococcus pneumonia***	**IV meropenem (42), Dexametheasone [4] Grommet insertion, rehabilitation. Survived**	**Current case**
67 Female	Depressed sensorium + vomit	*Streptococcus pneumoniae*	IV ceftriaxone (42) and Dexamethasone [3] Survived	Jayendrakumar CI et al. 2017 [14]
66 Male	Fever, subtle psychomotor retardation	*Methicillin resistant staphylococcus aureus*	Vacomycin followed by linezolid (49) Dexamethasone [4]	Marinelli L et al. 2014 [8]
63 Male	Fever and unsteady Left homonymous hemianopia	*Staphloccocus intermedius*	Cefotaxime (42) Metronidazole (42) Extraventricular shunt Survived	Vajramani G 2007 [11]
62 Male	Fever and headache after trip to Japan	*Listeria monocytogenes*	Vancomycin + Ceftriaxone [8] followed by Ampicillin and Gentamicin Dexamethasone [3] Extra-ventricular drain Died	Ito H et al. 2008 [10]
55 Male	Fever, intermittent occipital headache	*Neisseria meningitidis (blood cultures only – LP not performed due to raised INR)*	Clarithromcyin and Tazocin followed by ceftriaxone (41) + Rifampicin [9] Survived	Gronthoud F et al. 2017 [13]
54 Female	Sudden onset bilateral hearing loss	*Streptococcus agalactiae*	IV Ceftriaxone, surgery for mitral valve repair due to endocartiditis Survived Permanent hearing loss	Ito T et al. 2019 [15]
49 Male	Headache, confusion, meningism	*Streptococcus acidominimus*	IV Ceftriaxone Survived	Shah GS, 2018 [17]
45 Male	Hearing loss and vestibular nerve disorder	*Streptococcus suis*	Ceftriaxone and Ampicillin 24 days	Yanase T 2018 [18]
39 Male	Fevers, fatigue, frontal headache, meningism	*Enterrococcus faecalis* *Escheria coli peptostreptococcus spp*	Ventricular Shunt Survived	Barloon TJ et al. 1990 [12]

Additional therapies to consider include anti-epileptics if evidence of seizures. Intravenous steroids were used in four other cases but no clear evidence of improving outcome. *Lousard* refers to guidelines on starting steroids on obtaining a purulent LP to reduce complications of hearing loss [20]. The role of steroids is also of question as there was no evidence of raised intracranial pressure in this case or other case reports.

6. Conclusion

Much like meningitis, ventriculitis can result as a complication of otological disease; it can manifest as rapid neurological deterioration and is difficult to diagnose. There are no specific symptoms documented, apart from a noted absence of nuchal rigidity. Optimal work up includes radiological diagnosis with MRI and early lumbar puncture with CSF cultures for targeted, prolonged antimicrobial therapy. This is the first known reported case of ventriculitis associated with acute otitis media.

Ethical Statement for Solid State Ionics

Hereby, I Miriam Fahmy consciously assure that for the manuscript "Adult Primary Ventriculitis as a complication of acute otitis media: A comprehensive review of reported cases" the following is fulfilled:

1) This material is the authors' own original work, which has not been previously published elsewhere.

2) The paper is not currently being considered for publication elsewhere.

3) The paper reflects the authors' own research and analysis in a truthful and complete manner.

4) The paper properly credits the meaningful contributions of co-authors and co-researchers.

5) The results are appropriately placed in the context of prior and existing research.

6) All sources used are properly disclosed (correct citation). Literally copying of text must be indicated as such by using quotation marks and giving proper reference.

7) All authors have been personally and actively involved in substantial work leading to the paper, and will take public responsibility for its content.

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I agree with the above statements and declare that this submission follows the policies of Solid State Ionics as outlined in the Guide for Authors and in the Ethical Statement.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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