Atlanto-occipital septic arthritis complicating recurrent otitis media.

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ABSTRACT

Otitis media is known to have a number of complications. We present the first reported case of atlanto-occipital septic arthritis as a complication of *Streptococcus pneumoniae* otitis media in an eight month old boy.

INTRODUCTION

Acute otitis media is usually a benign condition. However, it may be associated with a number of complications, most commonly conductive hearing loss, but also by more serious infective conditions such as mastoiditis. Mastoiditis itself may be complicated by a number of infective conditions including labyrinthitis, septic thrombophlebitis, meningitis, intracranial abscess and central skull base osteomyelitis[5,1]. Spread of infection can occur through hematogenous, mucosal, bone or venous pathways . The most common bacterial pathogen in acute otitis media is *S. pneumoniae* accounting for 40% of all infections.

Septic arthritis of the atlanto-occipital joint is exceedingly uncommon and has not been previously described in children. We describe an 8-month-old boy who presented with fever and neck pain on a background of recurrent otitis media. Magnetic resonance imaging (MRI) of the head and neck was in keeping with atlanto-occipital septic arthritis and a right skull base collection. Blood cultures were positive for *S. pneumoniae*. He was treated with intravenous ticarcillin potassium-clavulanate for a total of seven weeks with complete resolution.

CASE REPORT

A previously well, 8-month-old boy was admitted to hospital with five days of fever, irritability and neck stiffness. He had completed the recommended schedule of immunizations for his age which included three doses of the seven valent conjugate pneumococcal vaccine. Four weeks prior to admission, he was seen by his local doctor who diagnosed acute otitis media and he received seven days of oral amoxicillin with complete resolution. Five days prior to admission he developed fever and was diagnosed clinically as having acute otitis media but was not prescribed antibiotics. He again presented to hospital two days prior to admission with ongoing irritability and fever measured at 39°C. He was diagnosed with bilateral otitis media and discharged without antibiotic therapy. One day prior to admission he re-presented hospital following a brief cyanotic episode. He was diagnosed with bronchiolitis, and was again discharged for planned review the next day.

On the day of admission he re-presented with ongoing fever and 12 hours of restricted neck movement. On examination his temperature was 38.8°C, pulse rate 162 per minute, respiratory rate 66 per minute. He was alert and became distressed when his neck was moved passively in any direction. Respiratory examination revealed slight suprasternal indrawing with no stridor. He had bilateral bulging, erythematous tympanic membranes.

Investigations revealed the following: haemoglobin 96g/L, platelet count $314 \times 10^9/L$, white cell count $10.6 \times 10^9/L$, lymphocytes $2.27 \times 10^9/L$, neutrophils $7.40 \times 10^9/L$ and C

1 reactive protein 177mg/L (normal value < 8mg/L). A lateral neck radiograph was
2 performed which demonstrated prevertebral soft tissue swelling.

3

4 An initial diagnosis was made of retropharyngeal abscess. Intravenous flucloxacillin,
5 cefotaxime and metronidazole were commenced. The patient remained febrile and
6 tachycardic with ongoing neck pain during the first twenty-four hours following
7 admission and the antibiotics were changed to intravenous ticarcillin potassium
8 clavulanate . A lumbar puncture performed after 24 hours revealed 1×10^6 /L
9 polymorphonuclear cells and $28\,000 \times 10^6$ /L red blood cells, with no bacterial growth
10 on culture. On the second day of his admission, a flexible laryngoscopy was
11 performed which did not demonstrate any bulging of the posterior pharyngeal wall,
12 therefore making a diagnosis of retropharyngeal abscess unlikely.

13

14 By day three of admission blood cultures taken at admission grew *S. pneumoniae*
15 (serotype 3) which was susceptible to penicillin (minimum inhibitory concentration
16 0.016mg/L). Lack of clinical improvement instigated an MRI of the head and neck
17 which revealed findings consistent with atlanto-occipital septic arthritis as well as a
18 large rim enhancing collection within the suboccipital soft tissue (Figures 1 and 2).
19 The collection extended across the midline to the left side of the occiput and also
20 insinuated anteriorly into the right and left atlantooccipital joint spaces. Drainage of
21 the skull base collection was attempted, however there was no fluid aspirated.
22 Bilateral tympanostomy tubes were inserted. The fluid drained by myringotomy grew
23 *S. pneumoniae* (serotype not available).

24

25 A repeat MRI seven days after admission showed marginal improvement in the skull

base collection but persistent involvement of the atlanto-occipital joint. The child was discharged home after insertion of a peripherally inserted central catheter and intravenous ticarcillin potassium-clavulanate was given for seven weeks in total. Sixteen days after admission the C reactive protein had normalized to 8 mg/L, white cell count $7.3 \times 10^9/L$ and neutrophils $2.0 \times 10^9/L$. After one month, a repeat MRI showed marked improvement. There was complete clinical recovery at four months.

DISCUSSION

We believe this to be the first reported case of atlanto-occipital septic arthritis as a complication of otitis media and we propose that septic arthritis of the atlanto-occipital joint be added to the list of known complications of otitis media.

Septic arthritis of the atlanto-occipital joint has not previously been described in children. However, central skull base osteomyelitis as a complication of otitis media has been described in both children and adults[2,1]. A 3-year-old girl with a previous history of cleft lip and palate reconstruction developed skull base osteomyelitis as a complication of mastoiditis[2]. Osteomyelitis was diagnosed on CT and bone scan one week after mastoidectomy. Bacterial cultures were positive for *S. pneumoniae*. She had complete clinical resolution. Septic arthritis as a complication of otitis media at other sites has also been described including septic arthritis of the shoulder joint due to *Pseudomonas aeruginosa* in a 23-month-old girl[3].

We postulate that the most likely pathogenesis of atlanto-occipital septic arthritis in

1 our patient was due to direct spread via the deep cervical fascial plane from the
2 tympanic cavity. This hypothesis is supported by the MRI findings. Both the
3 lymphatic and venous drainage of the tympanic cavity communicate with the deep
4 cervical fascial plane. There is a structural deficiency in the capsule of the atlanto-
5 occipital joint which allows the entry of the vertebral artery and which may allow
6 spread of the infection. Contiguous spread from the posterior air cells of the mastoid
7 to the occipital bone leading to osteomyelitis of the calvarium can also occur,
8 however in our patient there was no enhancement of the mastoid air cells on MRI nor
9 was there radiological evidence of osteomyelitis so this route is less likely.

10

11 Serotype 3 of *S. pneumoniae* was isolated in the blood culture in the case described.
12 Serotype 3 is not included in the seven valent pneumococcal conjugate vaccine of
13 which this patient had received three doses. However, serotype 3 is included in the 13
14 valent pneumococcal conjugate vaccine which replaced the seven valent vaccine in
15 the Australian schedule in July 2011[4].

16

17 Recommendations for treatment of septic arthritis vary from two to six weeks of
18 antibiotic therapy with the duration of the parenteral component of treatment varying
19 from only a few days to the entire six weeks[2]. Given the anatomical location, the
20 patient's age and the possibility of polymicrobial infection we elected to treat with a
21 prolonged course of intravenous ticarcillin potassium clavulanate. Haematological
22 side effects were monitored for by weekly full blood count which remained normal,
23 and the patient made a full recovery.

24

25

1 **CONFLICT OF INTEREST**

2 The authors declare that there are no conflicts of interest.

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Figures

Figure 1:

Figure 1a & 1b: Sagittal and axial T2 fat saturated images of the head and upper cervical region demonstrating the irregular fluid collection posteriorly adjacent to the right occipital bone (Arrow A). Fluid insinuates anteriorly into both atlantooccipital joints (Arrow B). Extensive bilateral high signal oedema is demonstrated extending through the muscle planes of the neck bilaterally (Arrow C).

Figure 2:

Figure 2a & 2b: Sagittal and axial post contrast fat saturated images demonstrate the large irregular low fluid signal rim enhancing collection adjacent to the right occipital bone (Arrow A). The collection is seen to extend anteriorly into the atlantooccipital articulations bilaterally (Arrow B).

Response to comments from reviewers at the Pediatric Infectious Diseases Journal

Our response to the reviewer comments are included in italics.

Reviewer #1:

Title.

-The title is too long and should be shortened to "Atlanto-occipital septic arthritis complicating recurrent otitis media".

We have shortened the title.

Abstract.

-8 month old boy sufficient, ½ month not needed.

Agree. We have changed this.

Background.

-First paragraph, most of the complications listed are of mastoiditis, which complicates otitis media. This should be more clearly stated.

We have changed this so that it is more accurate.

-A comment on the mechanism of how the spread of infection can occur which should include hematogenous, mucosal pathways and through bone (venous channels or bony destruction).

We have included a comment on the mechanism.

-Occipital osteomyelitis should be replaced with central skull base osteomyelitis.

We have made this change throughout the manuscript.

-Also a statement concerning the incidence of complications, increasing or decreasing, considering there is a clinical push to treat these children conservatively without antibiotics.

We have made a comment regarding the "clinical push" to manage children with AOM. Regarding the incidence of complications, this is difficult to assess as the studies conducted thus far are inconclusive. We initially made a change to more accurately reflect this in the article, however in the current version we have cut this out to reduce the word count and to make the report more concise.

-The second and third paragraphs should be merged.

Done.

Case report.

-Page 5 - 12th line, comma after fever.

Done

-37 th line paragraph is a repeat statement (1st and 3rd).

Removed.

-The 29th line beginning "The next day" should be joined to the next paragraph and it

seemslke this is the day of admission, which should be said.

We have changed this so that it is even clearer: "On the day of admission he represented with ongoing fever and 12 hours of restricted neck movement."

-Page 6. The first 2 paragraphs should be joined.

Done.

-If a diagnosis of RTP abscess was made, why was a CT not obtained or a repeat lateral radiograph or fluoroscopic evaluation? Why was a flexible laryngoscopy performed, should be clearly stated with pertinent negatives.

This section now reads: "On the second day of his admission, a flexible laryngoscopy was performed which did not demonstrate any bulging of the posterior pharyngeal wall, therefore making a diagnosis of retropharyngeal abscess unlikely. A computed tomography (CT) scan of the neck was not performed to avoid further irradiation."

-Second 2 paragraphs should be joined Page 7.

Done.

-Overall, the sequence of events is difficult to follow. The report should stick to the traditional days prior and after admission. The timing of the initial MR is unclear?

We have made a number of changes to this section of the manuscript, particularly by "sign-posting" the sequence of events by reference to the day of admission and we now believe it is clearer.

-Day 3 of admission. Line 12 changes should be changes to findings. The rim enhancing fluid collection is within the suboccipital soft tissue and this is more accurate than adjacent to the occipital bone.

Done.

-Last 2 paragraphs should be joined.

Done.

-Line 53 - More concise language needed - After one month, the MRI showed marked improvement with complete clinical recovery at 4 months.

"After one month, a repeat MRI showed marked improvement and there was complete clinical recovery at four months."

Discussion.

-It should be mentioned that other cases of septic arthritis have been reported, which includes direct extension to the temporal mandibular joint as well hematogenous dissemination to the shoulder.

We have included the case of hematogenous dissemination to the shoulder, and have commented on the occurrence of septic arthritis at other sites as a complication of otitis media.

-The described cases need to be more concise.

They are now more concise.

-Occipital osteomyelitis should be replaced with central skull base osteomyelitis (page 8, line19).

Done.

-Paul et al case. If there is destruction of the odontoid, would that not suggest involvement of the atlantodental joint?

We have removed this case from the discussion as it was not as relevant as we initially thought.

-Page 9. Second paragraph, concerning the statement no radiologic evidence of mastoiditis (line 48), how was this determined?

This has been clarified.

-The last paragraph is out of place and is merely a statement without relevance given.

We disagree. We think that the expanded serotype inclusion in the 13 valent vaccine is relevant to the case.

Reviewer #2:

This is a case report describing an infant with concurrent Strep pneumoniae otitis media and atlanto-occipital septic arthritis.

Major comments:

-There are a few studies of suppurative arthritis that mention otitis media as a concurrent infection (Welkon et al. Pediatr Infect Dis J 1986;5:669-, Goldenberg & Cohen Am J Med 1976;60:369- and Lejman et al. J Pediatr Orthop 1995;15:172-). These references should be included in the discussion of this case report.

We initially made changes to include these studies, but to keep the discussion concise we have mentioned the case of shoulder septic arthritis and that there are other reports of septic arthritis in other sites.

-The infant in the present study had lung infection as well prior to the diagnosis of septicemia and septic arthritis, so we do not know whether or not the middle ear was the source of this infection. Moreover, the speculation of the authors that the infection spread from the middle ear to the atlanto-occipital joint directly remains unknown as drainage of the occipital region yielded no fluid. Therefore, I would be more cautious in describing the causal relationship between the otitis media and septic arthritis.

Other than the one presentation with respiratory distress the patient did not display further signs of a chest lung infection. Therefore we are uncertain as to the accuracy of the initial diagnosis made of bronchiolitis. We believe the evidence points more towards otitis media as the primary diagnosis with septic arthritis as a complication. The slight respiratory indrawing on the day of admission may have been secondary to pyrexia given that the chest examination remained otherwise normal throughout admission.

Minor comments:

- the visits prior to admission are poorly described (which hospital, what kind of a doctor saw the patient- general practitioner, pediatrician, ENT specialist - symptoms, signs, possible lab exams, diagnoses)

We have now improved and streamlined the case history so that it is clearer (see responses to reviewer 1 above).

- choose either fig 1 or fig 2

We feel that all the figures provide valuable information because the combination of the T2 fat saturated images and the T1 post contrast images best demonstrate the pathology.

If three is the maximum we would suggest omission of the sagittal T2 fat saturated image (Figure 1a), and if only 2 images were allowed we would suggest further omission of Figure 1b.