CASE REPORT

# Skull Base Osteomyelitis Caused by *Achromobacter xylosoxidans*

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**Abstract**:

*Achromobacter xylosoxidans* is an aerobic, Gram-negative, non-lactose fermenting rod which inhabits soil and aquatic environments. It is uncommonly encountered in human infections. When it occurs, it is associated with nosocomial infections and infection in immunocompromised individuals. Awareness of its infectious potential is important as it is multi-drug resistant and can lead to increased mortality rates. We describe a case of skull base osteomyelitis secondary to otitis media caused by *Achromobacter xylosoxidans* in a diabetic patient. He responded to combination treatment with carbapenem and quinolone along with blood sugar level control. Both, the clinical disorder and the causative organism are exceptional. To the best of our knowledge, no case akin to this has been reported previously in literature.

**Keywords**: *Achromobacter xylosoxidans*, diabetes mellitus, middle ear, skull base osteomyelitis

## Introduction:

*Achromobacter xylosoxidans* is an aerobic Gram-negative, motile, non-lactose fermenting bacillus which is oxidase and catalase positive [1-2]. It occupies soil and aquatic environments like well water, and swimming pools in the community and intravenous fluids, humidifier water, saline or haemodialysis fluid in hospitals. It rarely causes human infections and its disease spectrum includes iatrogenic infections and outbreaks and primarily involves immunocompromised individuals [3-4]. It has occasionally been isolated from human gastrointestinal tract and middle ear [4]. Its scope of infection includes primary bacteraemia, catheter-related blood stream infection, endocarditis, otitis and pneumonia but infrequently involves temporal bone [4-5]. Skull Base Osteomyelitis (SBO) is a rare occurrence mainly involving the temporal and sphenoidal bone. It generally presents as a complication of Malignant Otitis Externa (MOE) usually caused by *Pseudomonas aeruginosa* infection [6]. We describe a case of SBO caused due to spread of middle ear infection which is an unusual clinical entity as much as the precipitating organism *A. xylosoxidans* and came to light due to knowledge of this possibility.

## Case Report:

A 72-year old male presented in our institute with complaints of right-sided headache for two days and change in voice, diplopia on right gaze and persistent right ear discharge for three months. There was no history of fever, vomiting, ear pain, trauma, swimming or exposure to well water. He is a known case of long-standing diabetes mellitus and hypertension on oral medications for both ailments. Three months ago, he was admitted in a private hospital for similar complaints along with neck pain and uncontrolled blood sugar levels. He gave history of right sided ear discharge on and off

and bilateral hearing loss for past fifteen years. He had undergone right ear surgery for the same fifteen years back in our institute. On examination he was conscious, well oriented, a febrile and demonstrated diplopia on right lateral gaze. Neurological examination revealed right lateral rectus palsy and a well-compensated right vocal cord palsy. Other signs like finger counting, light perception, facial symmetry, swallowing, shoulder shrugging and tongue movement were normal. Ear Nose Throat (ENT) examination revealed right sided large central tympanic membrane perforation with slightly inflamed margins and minimal discharge. Weber's test was lateralised to right. Rinne's test was negative on right and equal on left. Pure tone audiometry revealed right moderate mixed and left moderate sensorineural hearing loss. On his previous hospital admission three months ago, C-reactive Protein (CRP) was 64 mg/Land Erythrocyte Sedimentation Rate (ESR) was 100 mm/hour. Gram stain of ear discharge did not show organisms and grew *Pseudomonas aeruginosa*. Magnetic Resonance Imaging (MRI) was suggestive of skull base osteomyelitis with marrow signal abnormality and erosion in right Petrous apex, right half of clivus involving region of Dorello's canal, margins of Jugular foramen and Hypoglossal canal. There was enhancing soft tissue bulge in prevertebral region involving longus capitis muscle and extending to right nasopharynx and fossa of Rossenmullar area. Biopsy from this area was suggestive of infection without evidence of malignancy or granuloma. GeneXpert test of that tissue was negative. Patient was treated with meropenem and piperacillin-tazobactam as per the antibiotic susceptibility report for four weeks and discharged on clinical improvement. On current admission, his ESR was 25 mm/hour and CRP was 1 mg/L. Fasting and post-prandial blood glucose levels were 198 mg% and 122 mg% respectively, HbA1c was 11.1% and other blood investigations were normal. Repeat MRI with contrast showed a large area of persistent altered signal intensity involving the same areas as the previous MRI and additionally involving right petrous temporal bone (Fig. 1). Post contrast images were suggestive of abscess formation and right mastoiditis. Right ear discharge collected from Eustachian tube area of middle ear was received for aerobic culture and antibiotic susceptibility testing. Gram stain of the sample showed few pus cells and slender Gram-negative bacilli (Fig. 2). Ziehl-Neelsen stain was negative. After 24 hours of incubation at $37^0$C, pure growth of non-lactose fermenting, circular colonies with round margins was seen on MacConkey agar (Fig. 3) and smooth, glistening, entire, non-haemolytic, non-pigmented, odourless colonies on Sheep Blood agar. Colonies on nutrient agar were non-pigmented. Gram stain of the growth showed slender Gram-negative bacilli which were motile and oxidase and catalase positive. It was identified as *Achromobacter xylosoxidans* on Vitek 2/ Compact automated system (Biomeriux, India). It was susceptible to imipenem and colistin, showed intermediate susceptibility to meropenem and levofloxacin and resistant to ciprofloxacin, amikacin, gentamicin, ceftriaxone, cefepime, ceftazidime, piperacillin-tazobactam, cefoperazone-sulbactam, ticarcillin-clavulanic acid and trimethoprim-sulfamethoxazole. He was given intravenous meropenem 1 gram twice a day and levofloxacin 500 mg once a day intravenously along with insulin and oral antidiabetic drugs to which he responded clinically. He was discharged

after six weeks on oral levofloxacin and trimethoprim-sulfamethoxazole. Follow-up MRI after three months showed that the abscess was resolving.

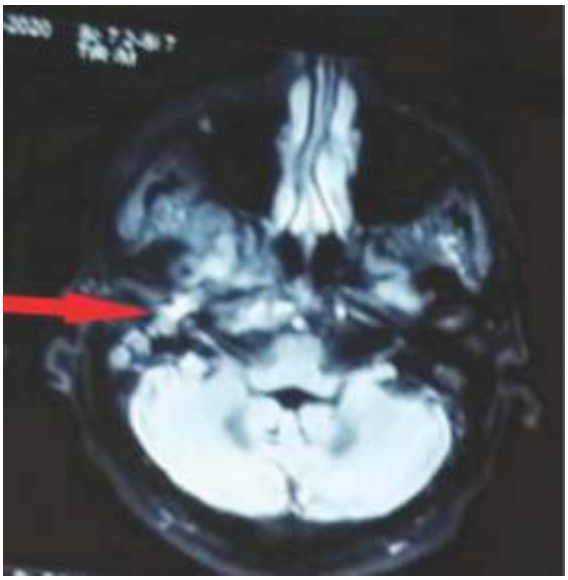

**Fig. 1: MRI Scan showing Involvement of Right Petrous Temporal Bone**

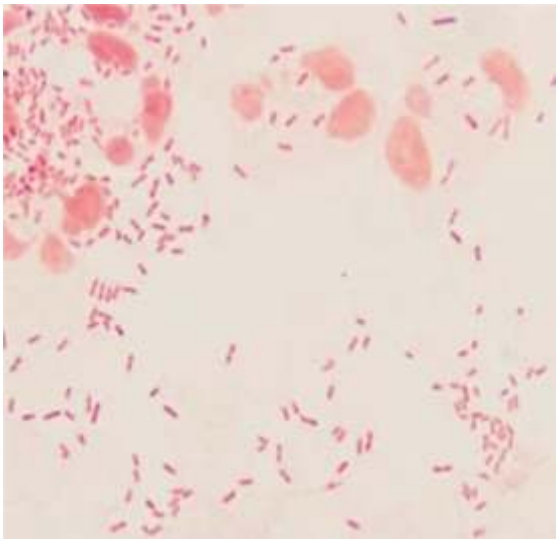

**Fig. 2: Gram Stain showing Pus Cells and Gram-negative Bacilli**

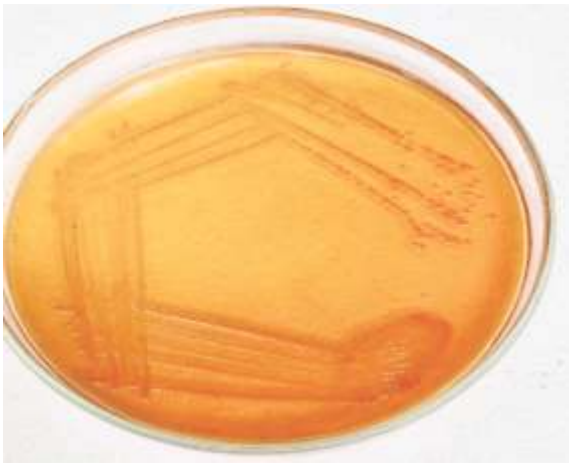

**Fig. 3: Colonies of *A. xylosoxidans* on MacConkey Agar**

**Discussion**:

*A. xylosoxidans* occupies aquatic environments in community and hospitals. It can also survive in commonly used disinfectants like chlorhexidine, alcohol and quaternary ammonium compounds [7]. It is probably part of the indigenous microbiota of the ear [8]. It appeared that our patient had right middle ear infection as seen on ENT examination and presence of pus cells and Gram-negative bacilli in the discharge which subsequently grew *A. xylosoxidans*. He must have acquired this infection from hospital, community or while daily bathing which was treated partially and so recurred. His high blood sugar levels induced its spread to the skull base. Cranial osteomyelitis is an unusual clinical condition which can involve cranial vault or skull base. SBO is a catastrophic, complex clinal condition involving the temporal and sphenoidal bone which is generally a direct complication of otogenic infection [6]. The commonest cause of SBO is MOE caused by *Pseudomonas aeruginosa* especially in poorly controlled diabetes mellitus

[6]. But in this case, infection lay in the middle ear and not the EAC which makes this case special. Previous culture report of ear discharge revealed growth of *P. aeruginosa*. But it is possible that the causative organism could have been replaced by *A. xylosoxidans* over a period of time as seen by the current pure growth and cultural characteristics of this isolate which were dissimilar to that of the former. Another probability could be that both these organisms must have grown then but *Pseudomonas* being a known pathogen of the ear was reported whereas *Achromobacter* was disregarded as a contaminant. *A. xylosoxidans* is known to be multi-drug resistant and hence is required to be adequately identified. In this case too, patient's quinolone antibiotic was changed from ciprofloxacin to levofloxacin as per his current antibiotic susceptibility report. Cranial nerve involvement is seen in SBO as the disease progresses commonly and involves facial nerve followed by ninth, tenth and eleventh cranial nerves and jugular foramen [6]. Our patient also presented with right lateral rectus palsy and right vocal cord palsy indicating involvement of the sixth and tenth cranial nerve. An uncommon variant of SBO is atypical SBO which manifests as headache, atypical facial pain and cranial nerve palsies. It may not present with signs of infection like fever, leucocytosis and elevated Erythrocyte Sedimentation Rate (ESR) [6]. Similar findings in our case were noted except ESR which was raised earlier but decreased due to treatment. But atypical SBO shows sphenoidal and occipital involvement along with Clivus rather than temporal bone as seen in typical SBO [6]. All these findings evocative of probable atypical SBO were exhibited by our patient except MRI finding which showed involvement of temporal bone along with clivus. This further progressed to abscess due to spread of his middle ear infection predisposed by his immunosuppressed status due to prevailing diabetes mellitus. On the whole, this scenario is out of the ordinary. Irrespective of type, treatment modality of bacterial SBO comprises of long duration course of broad-spectrum antibiotics ranging from three to six months [6]. Our isolate was resistant to most of the antibiotics except carbapenems and quinolones and was treated accordingly along with measures for glycaemic control. This organism has an abundance of intrinsic resistant mechanisms with a wide-reaching resistance pattern involving many drugs and shows a similar susceptibility pattern as ours [8-10].

**Conclusion**:

Our case of SBO caused by *A. xylosoxidans* as a complication of middle ear infection in a diabetic patient is an unusual presentation. This case came to light due to heightened awareness of the infectious potential of organism and was not disregarded as a contaminant. Timely and accurate identification from the laboratory and the clinician's expertise in managing the case averted the patient's condition from intensifying.

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