Rare Case Report: Serratia Liquefaciens and Enterococcus Species as a Cause of Otogenic Brain Abscess Secondary to Active Epithelial Chronic Suppurative Otitis Media (CSOM) in a 12-Year Old Male Patient with Hemiparesis

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Abstract

Developing countries face the problem of otogenic intracranial complications especially in paediatric population because of poor hygiene, malnutrition and low immunity. Active epithelial type Chronic Suppurative Otitis Media (CSOM) can be the cause of potentially fatal complications, one of these is otogenic brain abscess. Having an estimated annual incidence to be approximately 0.5 per 100,000 children, it is associated with high mortality.

We describe a unique case of otogenic brain abscess secondary to Active epithelial CSOM being neglected for two years and finding an unexpected foreign body in the right ear canal in a 12-year-old boy. The consultation was sought when the headache became intense and hemiparesis appeared. Our case is unique regarding both the clinical presentation and the causative organism. The patient underwent craniotomy, with elective capsulotomy, drainage of abscess and placement of a ventriculoperitoneal shunt. This was followed by a modified radical mastoidectomy two weeks later and an uneventful recovery.

Keywords: *Active epithelial (Previous Atticoantral) CSOM, otogenic brain abscess, otogenic hydrocephalus, serratia liquefaciens, intracranial otogenic complications.*

INTRODUCTION

Developing countries still face the problem of otogenic intracranial complications; one serious and life-threatening being otogenic brain abscess associated with Active epithelial type CSOM [1].

Paediatric brain abscesses have an estimated annual incidence to be approximately 0.5 per 100,000 children [2, 3]. The classic triad of brain abscesses *i.e.* fever, headache, and focal neurologic deficits, are found in only 20% of patients according to one study [4, 5].

Here we report a unique case of otogenic brain abscess secondary to chronic mastoiditis and Active epithelial CSOM. Complications developed due to late presentation by the patient and little knowledge by the patient and his attendants about the seriousness of the situation as the condition had been neglected for two years. The patient only complained of two days history, when the symptoms especially headache became intense and hemiparesis appeared. Our case is also unique regarding the causative organism, as this condition is most commonly reported to be caused by Proteus, streptococcus milleri group [6].

CASE REPORT

A twelve-year-old male patient was admitted to the Paediatric department of Ayub Teaching Hospital (ATH) on February 08, 2022. The attendants gave a two-day history of fever and decreased appetite, an agonizing generalized headache and right-sided weakness of the body since one day. According to the attendant (father), the onset of symptoms was short, he was well two days before when he gradually developed low-grade continuous fever (undocumented). The fever was associated with decreased appetite but not with chills or shivering. The next day he developed a worsening headache and three episodes of vomiting. Vomiting was non-bilious, non-bloody and not associated with food intake. The patient took three tablets of paracetamol and 1 tbsp of syrup Pizotifen to ease his continuous headache and vomiting. He was a developmentally normal child, a student of grade three, fully vaccinated, without a history of any hospitalization, transfusions or drug allergies. His birth history was normal. He had a history of ear discharge on and off for an unknown duration for which he took multiple treatments from the locality but the discharge hadn't recurred for the last five months. History of ear infection was only disclosed

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upon direct questioning, after finding evidence on brain imaging a few days later after admission. There was no significant family history of any illness. He had three sisters and belonged to poor socioeconomic status.

On general physical examination, the patient looked pale. GCS was 15/15 with altered mental status, though it seemed more due to the headache he was experiencing. The patient looked anxious and was in obvious pain and discomfort. On systemic examination, heart rate was regular at 70 beats per minute; Blood pressure was in the normal range for his age *i.e.* 80/50 mmHg. The patient was febrile with an axillary temperature was 99.5ºC. There was crusted right ear wax / dried-up discharge from the right ear. Fundoscopic examination of the eyes showed bilateral blurred temporal disc margins. He was started on IV mannitol and oral acetazolamide. On neurological examination he was drowsy, irritable, with signs of meningeal irritation; Neck rigidity, Kerning's and Brudezinski's signs were positive, reflexes were brisk and plantars were upgoing. Power was slightly reduced on the right side in both upper and lower limbs. Cranial nerve examination and cerebellar signs were negative. All other systemic examinations came as unremarkable.

A differential diagnosis of intracranial infection (Septic meningitis), brain abscess, tuberculous meningitis and the intracranial space-occupying lesion was formulated. Accordingly, workup was started.

Initially considering him a case of intracranial infection based on the short duration of symptoms and examination, treatment was started empirically as IV ceftriaxone, vancomycin and acyclovir in accordance with the clinical presentation and ward protocols. Intravenous steroids were added to reduce inflammation. Initial labs showed a TLC count of 16000, CRP levels 16 (positive), and Hb 12.9 g/dl, other labs that included renal profile, liver profile and electrolytes were in normal reference ranges.

The patient showed improvement after two days of treatment but the headache persisted. Cerebrospinal fluid (CSF) was sent for routine analysis and when the CSF routine analysis report was received, it showed a rise in total cell counts with predominant lymphocytes and an increase in CSF total proteins. CSF R/E showed low sugar of 50mg/dl, raised protein of 158mg/dl, RBC 40cells/mm and raised WBC 400cells/mm with neutrophil predominance of 75%. Aciclovir was discontinued in light of the CSF findings.

After three days, the patient showed no signs of Improvement on the above-mentioned antibiotics. The antibiotics were reviewed and changed to Meropenem along with vancomycin, IV Flagyl was added. With no noticeable improvement in his condition by the 10th day of admission, it was decided to repeat the CSF R/E along with a CSF gene expert to rule out tuberculous meningitis (TBM). Repeat CSF showed no marked response, repeat CSF sugar was 47mg/dl, CSF proteins raised

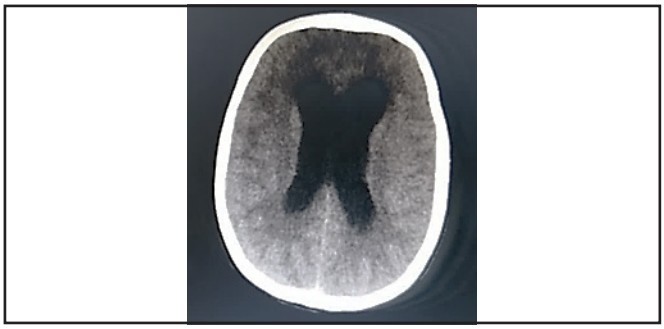


Fig. (1): First CT Brain imaging shows generalized cerebral oedema and ventricular dilatation with post-contrast enhancement.

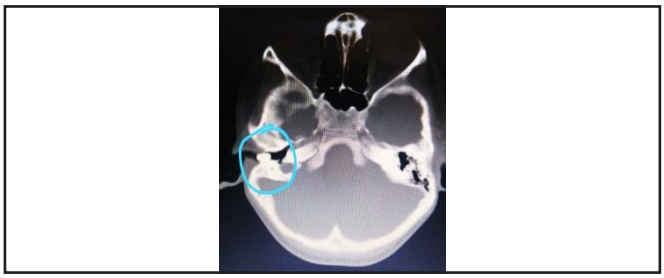


Fig. (2): Foreign body in the right ear canal (encircled).

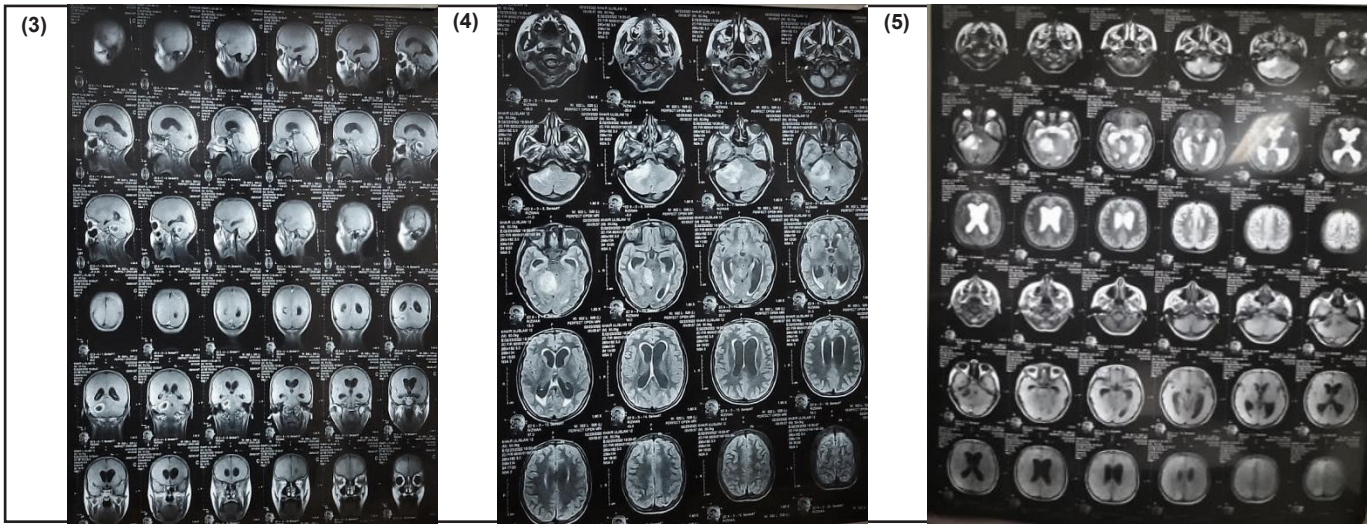
to 178mg/dl, RBC 1200 cells/mm and markedly raised WBC 4600cells/mm with the neutrophil predominance of 75%. CSF was sent for culture. Workup for tuberculosis *i.e.* CSF Gene expert, Chest X-ray, Montoux test came negative.

The first CT brain images came and revealed dilation of ventricles. A call was sent to a neurologist, and antituberculous medicines were started empirically.

In addition to dilatation of the lateral ventricle, the CT brain showed generalized brain edema and patchy post-contrast enhancement (**Fig. 1**). Also noted in the CT brain images were foreign body in the right ear canal and changes associated with chronic mastoiditis (**Fig. 2**). Keeping in view of suspicion of otogenic brain abscess an urgent brain MRI was planned. In response to direct questioning, he gave a history of intermittent copious ear discharge on the right side for the last two years.

While MRI was awaited, his condition deteriorated under the effect of a severe headache and his GCS dropped to 13/15. The patient also developed facial palsy; Meanwhile, ENT was consulted for the removal of a foreign body in the right ear. A detailed ENT examination and suctioning revealed three small pebbles in the patient's right ear canal. His GCS further deteriorated after suctioning. Upon inquiring on this matter the patient recalled inserting the pebbles himself two years prior at the age of 10 years. This history of the foreign body in the right ear was previously not mentioned.

Report of MRI brain revealed brain abscess measuring approximately 2 cm in craniocaudal, 2.2 cm in AP and 2.2cm in transverse dimension in the anterior aspect of right cerebellum causing mass effect and obstructive hydrocephalus (**Figs. 3-5**). A diagnosis of otogenic hydrocephalus and otogenic brain abscess was made.



Figs. (3-5): MRI of a brain on 15th day showing soft tissue and cystic component in the anterior aspect of right cerebellum causing obstructive hydrocephalus and mass effect.

The patient was shifted to the neurosurgical unit as his GCS was deteriorating and urgent extra ventricular drainage of CSF was done under Kocher's procedure to relieve rising intracranial pressure. He was then planned for elective capsulotomy after two days. Tracheostomy was done as the patient was being prepared for general anesthesia and surgery.

After the initial palliative measures, drainage of brain abscess was planned. His brain abscess was drained under general anaesthesia by craniotomy of the right side and pus was sent for culture and sensitivity. With 48 hours of incubation, histopathology showed the growth of numerous gram-positive cocci in chains (Enterococcus species) sensitive to chloramphenicol/ vancomycin and resistant to penicillin as well as occasional gram-negative bacilli (Serratia liquefaciens) sensitive to chloramphenicol, ceftriaxone and meropenem. It was mentioned that Serratia can develop resistance against 3rd generation cephalosporins within 3 or 4 days of treatment so a repeat culture should be sent. This is the first time that Serratia as a cause of otogenic brain abscess is being reported as Serratia unusually causes brain sepsis or infections in newborns. Repeat post-op CT Brain showed considerable improvement in the otogenic hydrocephalus.

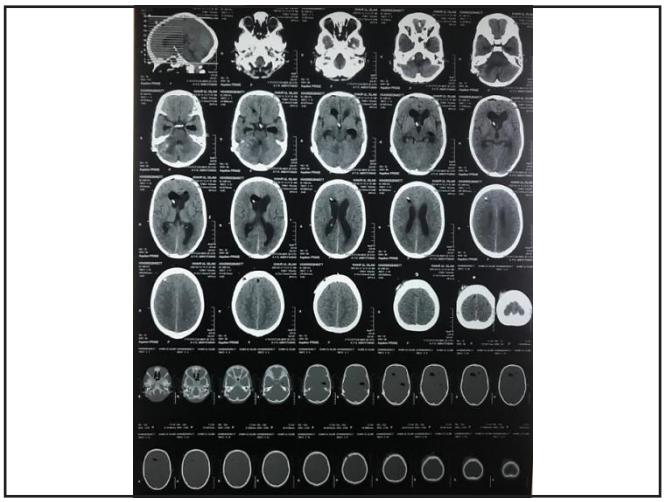


Fig. (6): CT brain with the improvement of hydrocephalus after post-op and in situ VP shunt.

The patient clinically showed improvement as well and his drain was removed following serial CT brain and replaced with the ventriculoperitoneal shunt (**Fig. 6**).

Next in the plan was to treat mastoiditis. After two weeks of recovery, the patient was admitted to the ENT ward in case of complicated CSOM for elective mastoidectomy. A pure tone Audiogram was done, results revealed right-sided mixed hearing loss. Modified radical mastoidectomy was done under G.A, An extensive cholesteatoma was noted in the mastoid and bone cavity. Dura and sigmoid sinus were exposed; Sino-dural angle cholesteatoma with the defect was noted as the most probable site for the intracranial spread of disease. The ossicular chain was intact. Meatoplasty was done, BIPP was kept in the mastoid combined cavity for 2-3 weeks and an incision

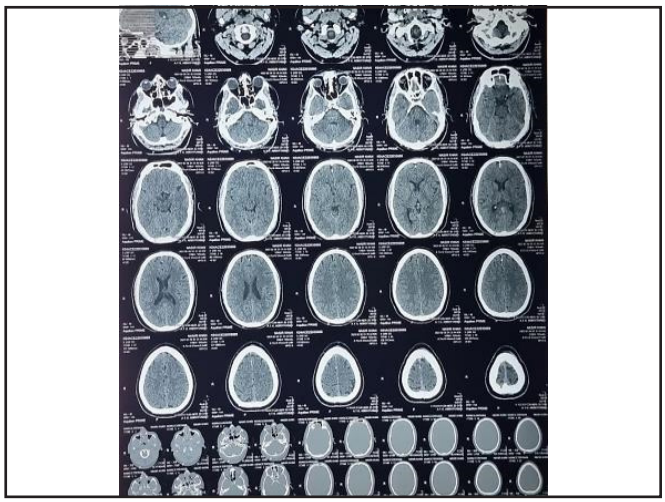


Fig. (7): Follow-up CT brain.

was closed in layers, the antiseptic dressing was done, recovery was uneventful and the patient recovered fully.

Upon the latest follow-up visit, a substantially improved CT brain showed a craniotomy defect noted in the right craniotomy bone with a small pneumocranium, minimal haemorrhage in the right occipital lobe 3cm away from the craniotomy site (**Fig. 7**). The patient was fully ambulant with a resolution of the presenting complaints.

DISCUSSION

CSOM is a chronic inflammation of the middle ear cleft, divided into tubotympanic and unsafe Active epithelial CSOM. The latter destroys the bones and leads to intracranial complications [7].

Affecting the poster superior part of the middle ear (attic, antrum, and posterior tympanum, mastoid) Active epithelial CSOM can lead to bone erosion and formation of cholesteatoma, with the risk of serious complications. Other associated pathological processes are osteitis, granulation tissue formation, ossicular necrosis and cholesterol granuloma. Common symptoms are foul-smelling ear discharge, hearing problems, giddiness and headache, *etc.*, and depend upon the severity of the disease. Due to the critical location of the temporal bone, mastoid and middle ear complex; the proximity of the facial nerve, and its separation from the middle and posterior cranial fossa by a thin bone, infections can extend intracranially [7, 8].

Still a major problem in developing countries, otological intracranial complications can be life-threatening. While small otogenic brain abscesses, which are less than 1.6 cm in size respond to treatment with antibiotics, and can be managed by medical therapy, treatment of larger otogenic brain abscess is immediate neurosurgical drainage after stabilizing the patient, followed by mastoidectomy to remove the source of infection. The ideal time for mastoidectomy is controversial. Murthy *et al.* affirmed neurosurgical drainage first, followed later by an ear operation. Singh and Maharaj and Mathews and Maurus recommended first neurosurgical drainage and later mastoidectomy, both in the same setting and in this order [8-10].

Our patient presented with facial palsy and hemiparesis which in children is considered an unusual complication of CSOM especially when the disease isn't diagnosed and treated timely and amply, thus causing the infection to spread intracranially [11].

The frequency of common complications along with aetiology is mentioned in texts [12]. Our case was unique both in terms of clinical presentation as well as aetiology.

Our patient also had otogenic hydrocephalus so prior to capsulotomy extra ventricular drainage was done or reduced pressure followed later on with proper placement of a ventricular peritoneal shunt. A similar case of CSOM with cholesteatoma was reported by Darmawan AB in a 14-year boy with complications of right hemiparesis after an otogenic brain abscess. The patient went through open craniotomy, drainage of the abscess, radical mastoidectomy and drug-sensitive intravenous antibiotics [11].

The organisms isolated from our patients were heavy growth of enterococcus species and light growth of serratia liquefaciens and this is a rare occurrence. Predominant organisms in one-third of cases of brain abscess are streptococci. Others include Pneumococcus, enterococcus and viridans with Staphylococcus accounting for 11% of cases, especially in penetrating injuries, gram-negative anaerobes are isolated in others. Whereas in neonates, serratia marcecens is an important cause of hospital-acquired infections, especially in neonatal intensive care units (NICUs), in addition, proteus mirabilis is common. Atypical organisms such as nocardia, mycobacterium, listeria and fungi are common in immune-compromised. As reported in other studies, in 27% of cases mixed growth occurs, especially in sinusitis or dental infection-associated cases. Sterile cultures are noted in ranges of 10–56% in the published literature [13-15].

Regarding outcome, as the intracranial abscess is a formidable entity and despite the advent of newer antibiotics and surgical strategies, the overall outcome and quality of life issues in brain abscess patients still remain a continuous challenge for the neurosurgical community [16].

When left untreated, brain abscess is deadly in 100% of cases. With timely intervention and treatment, the death rate can be reduced to 10% to 30%. The earlier the treatment is commenced, the better [17].

In one study the survival rate in children with temporal lobe abscesses was 90% and 75% in those with cerebellar abscesses, while among adults the survival rate was about 80% for both localizations [18, 19]. Fortunately with diagnosis soon after admission, collaboration among specialities (paediatrics, neurosurgery, neurology and ENT), and timely intervention our patient had a favourable outcome and was discharged in excellent health.

CONCLUSION

CSOM with its complications is not uncommon in developing countries such as Pakistan. High suspicion should be kept for rare presentations like hemiparesis, especially in those with a positive history and a risk of intracranial complications such as in our case, as delay in diagnosis can be fatal. Collaboration between paediatricians, neurosurgeons and otologists is needed for effective management and favourable outcome.

CONSENT FOR PUBLICATION

Written informed consent was taken from the patient's parents.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

ACKNOWLEDGEMENTS

Declared none.

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