Review Article

Sphingomonas paucimobilis — Related Central Nervous Infection : A Systematic Review

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Abstract

Background : Sphingomonas paucimobilis is a gram-negative bacterium that is an emerging human pathogen that causes both community-acquired and nosocomial infections. It can affect various sites, including the central nervous system. The prognosis is typically favourable, although it has been linked to a few severe events, including death.

Aims and Objectives : The discuss about all the cases of Sphingomonas paucimobilis infections of the Central Nervous System (CNS) in the general population that have been established by bacteriology.

Materials and Methods : *Data sources* - Pubmed, Cochrane library and Directory of Open Access Journals.

Eligibility Criteria - Studies mentioning CNS infections due to S paucimobilis in humans.

Synthesis methods - PRISMA

Results : There were 9 papers covering 12 patients enrolled, 4 patients of which were part of a study for which individual information were not available. Meningitis, which was largely community acquired and seen among immunocompetent adults, was the most prevalent diagnosis among the remaining 8 cases. All patients were treated with antibiotics with an average duration of 21 days. 3 developed complications out of which two expired, the remaining had good outcome.

Limitations : Small sample size and incomplete clinical details from one study.

Conclusion : Sphingomonas paucimobilis is an occasional human pathogen causing infection including CNS. Though most of the cases have favourable outcome but complications and death have been noted. The infection responds effectively to antibiotics, hence early identification would be lifesaving.

Key words : Sphingomonas paucimobilis, Meningitis, Cns Infection, Ventriculitis.

Sphingomonas paucimobilis is a gram-negative, non-fermentative, low motility bacillus that is an emerging human pathogen[1]. Though most infections caused by it have favourable prognosis few instances of fatal outcomes have been reported[2-4]. Despite causing mortality on a few occasions, little is known about its effect on the central nervous system.

The purpose of this study was to conduct a systematic review of Sphingomonas paucimobilis infections of the Central Nervous System (CNS) in the general population that have been established by bacteriology. A secondary goal was to characterise the clinical traits, diagnostic, treatment approaches, and outcomes of individuals with S Paucimobilis-related CNS infections.

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Editor's Comment :
- Sphingomonas paucimobilis is a rare but can potentially be a serious human pathogen.
- Early recognition and prompt antibiotic therapy are crucial, as timely treatment can lead to favorable outcomes and prevent complications or death.

Materials and Methods

Pubmed (https://www.ncbi.nlm.nih.gov/pubmed/), Cochrane Library (http://www.cochranelibrary.com/) databases and Directory of Open Access Journals (till October 10, 2023) were systematically searched for studies reporting Central Nervous System infection caused by Sphingomonas paucimobilis. We used the search phrases [(pseudomonas paucimobilis) or (Sphingomonas paucimobilis)] and [(meningitis) or (cns infection) or (central nervous system infection) or (ventriculitis) or (encephalitis)] to find clinical studies to review. The titles and abstracts of the retrieved publications were reviewed for eligibility. The following inclusion criteria were used to select studies for further analysis: (1) full-text publication/abstract reporting at

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least one Sphingomonas paucimobilis-related CNS disease, (2) enrolling human subjects and (3) In English language. Conference reports, remarks, and findings from studies using animals or cell lines were removed. Citations selected were included in the final analysis if these following data were available: documentation of CNS infection and confirmation of S paucimobilis detection by cultures.

All data were independently abstracted in triplicate by three investigators (DRR, NK, KK) according to the inclusion criteria. The first author's last name, the publication year, the age of the reported patients, their diagnosis, mode of acquiring the infection, their immune status, the complications encountered, the method of bacterial isolation, the bacterial antimicrobial susceptibility and resistance profiles, the initiated treatment, as well as the outcomes were all information that was retrieved from each publication. SPSS1 version 24 was used to do descriptive analysis on the extracted data (IBM, Armonk, NY, USA).

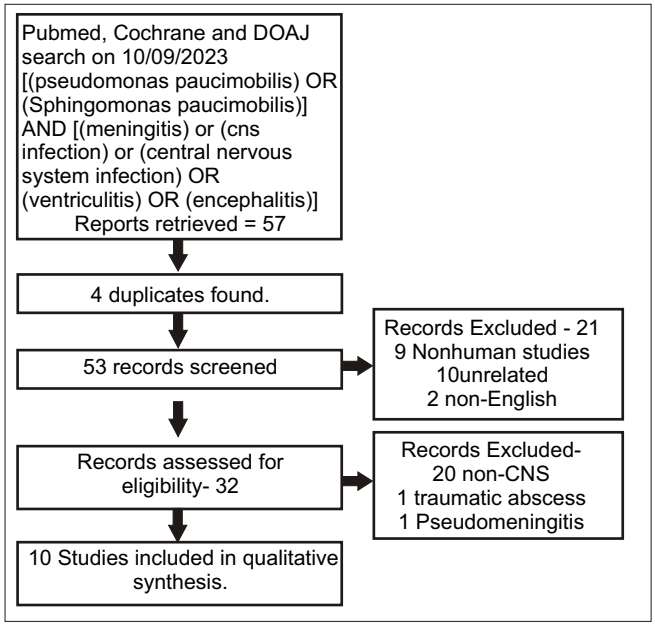

Fig 1 — PRISMA flow diagram of describing inclusion and exclusion criteria for experimental studies
DOAJ - Directory Of Open Access Journal,
CNS - Central Nervous System

Results

Our literature review yielded 17 publications from Pubmed, none from Cochrane library and 40 from DOAJ, of which 4 were duplicates hence removed. Out of these we excluded 21, as 9 were non-human studies, 10 were unrelated and 2 were in English language. Out of the remaining studies after eligibility checkup, 20 of them were found to be non-CNS infections, 1 case was a post traumatic abscess and 1 case was having pseudomeningitis hence were not included. The remaining 10 publications were further evaluated. A flow-diagram summarizing the literature research approach is shown in Fig 1. A triplicate screening was done and all the 9 were found to be meeting our inclusion criteria and hence were analysed for our systematic review.

We were able to find out a total of 13 cases of S paucimobilis infection of central nervous system. Bayram N, et al in his study form 2013, analysed the S paucimobilis infections among 24 children, out of which there was one case with CNS infection [5]. Complete details of this patient were not available. Similarly, Rohilla, et al in their study done in 2021 from a teaching hospital in India reported 4 cases of S paucimobilis isolated from CSF among a total of 49 isolates[6]. The details of each patient however were not available for this review. Of the other 8 cases the most frequent diagnosis was meningitis (7 patients, 88%, diagnosis was not specified for 1 case[3]) and only one case had associated ventriculitis.

All patients were adults with a mean age of 46 years (median age 45 years, range 30-66 years) and 4 of 8 were females. Majority of the patients were immuno-competent (6 patients, 75%), one article by Marincu N, et al did not mention about the immune status[7]. In 5 patients were noted to have underlying illness. The median duration of symptoms was 7 days (range of 1-21 days), Bolen, et al and Marincu, et al did not mention the duration of illness. In 7 cases were community acquired (88%), however Bolen, et al dint mention about the same[7,8].

The common method used to isolate the bacteria was conventional with Vitek 2 (50%) however one study each used MALDITOF[8] and BacT Alert[6] methods. One case had a simultaneous CNS infection with Mycobacterium tuberculosis, 7 rest all grew single organism from CSF.

Two patients had concomitant blood culture growths, one yielded S paucimobilis[9] and the other was listeria monocytogenes[10]. All the patients were treated with antibiotics and 2 of them received steroids. The average duration of antibiotics noted among those who survived was 21 days. The commonest complication noted was hydrocephalus (3) followed by ventriculitis and IVH (1 each). 4 patients underwent

surgical intervention, of which 3 were External Ventricular Drain (EVD)[3,4,10] and 2 Ventriculo-Peritoneal (VP) shunt[10,11]. Out of 9 patients, 6 had recovery among which one was recovering and was bedridden without neurological deficits10 however 2 patients did not survive[3,4].

Discussion

The genus Sphingomonas was described by Yabuchi, *et al* in 1900 and emended by Takeuchi et al in 1993, comprises of 12 species among which Sphingomonas paucimobilis is an occasional human pathogen, formerly known as CDC group IIk, biotype 1 then later on it was named as Pseudomonas paucimobilis in 1977[1,12,13]. In 1990, this organism received its own genus Sphingomonas based on the type of strain of the genus. It shares similar biochemical properties, fatty acid content with flavobacterium.

The first case of P paucimobilis was reported in 1979 in a sailor with leg ulcer and was isolated in pure culture. Subsequently many more infections with Sphingomonas have been reported[6,14,15].

Sphingomonas can be divided into four phylogenetic groups each representing separate genus these are, Sphingobium, Novosphingobium, Sphingophyxis along with Sphingomonas[1].

There are three species of Sphingomonas - Sphingomonas parapaucimobilis[1], Sphingomonas mucosisimma[16] and Sphingomonas adhesive[17] which have been reported to cause infections in humans.

Sphingomonas paucimobilis is a straight gram negative, non-sporing rods with single polar flagellum. (Fig 2)[1,13,18]. However when incubated at 18 to 22°C, it is motile on wet mount and motility medium, and is non-motile in 37°C hence the name paucimobilis due to difficulty in demonstrated motility in laboratory[1,18]. It lacks lipopolysaccharide in its outer capsule[12]. Instead it possesses two different kinds of Sphingolipids (hence the name Sphingomonas). Sphingolipids are unique sphingoglycolipid with long chain base di-hydrosphingosin, ubiqioune 10 Q-10 and hydromyristic acid 2-OH C 14:0 and absence of 3- hydroxy fatty acids (Fig 3)[19].

These are oxidase and catalase positive. On Sheep blood agar it grows as deep yellow colony with optimum growth seen at 30°C in 5 % CO_2 or ambient air, also grows in 37°C but do not grow at 42°C. It

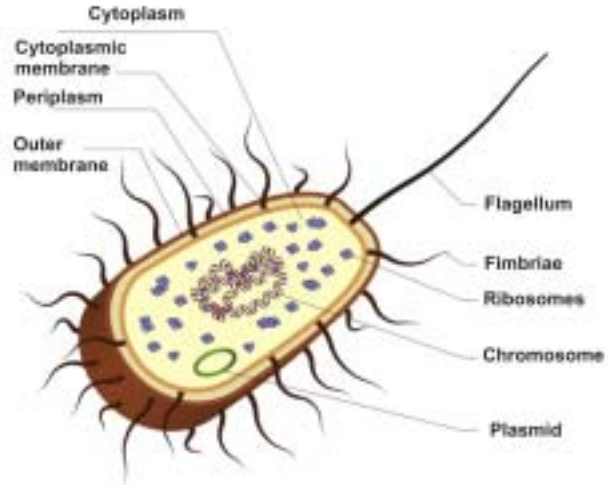

Fig 2 — Microscopic representative image of Sphingomonas paucimobilis

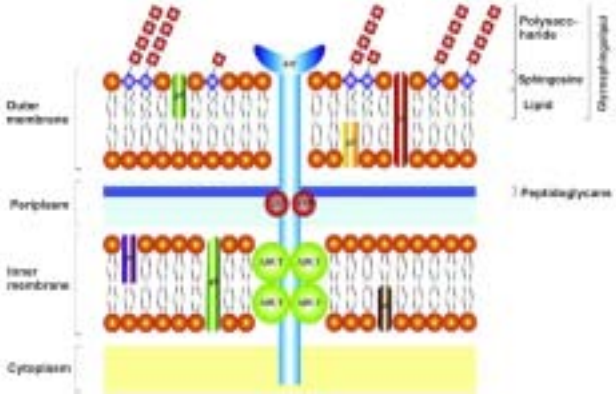

Fig 3 — Bacterial cell wall

does not grow on MacConkey agar (90% does not grow, 10% grow as non-Lactose Fermenters). It utilizes glucose, xylose and sucrose oxidatively, strongly esculin hydrolysis positive, urease negative and indole negative, produces a zone a inhibition around vancomycin disk places in BAP. It is susceptible to polymyxin B, which differentiates it from Sphingobacterium. Sphingomonas paucimobilis have been isolated from blood, CSF, urine, wounds, vagina, cervix and hospital environment. Most strains are susceptible to tetracycline, chloramphenicol, trimethoprim-sulfamethoxazole and amino-glycosides[1,13,18].

The organism is mostly acquired from the community[2], but it has also been linked to nosocomial infections and has been isolated from hospital equipment, water source and devices[6,15].

Only a few case reports and short case series have

described S paucimobilis's pathogenicity in humans, and one systematic review linked it to infections of the bones and soft tissues[15]. The organism is associated with infections of various sites[6] with a predilection to bone and soft tissue and only 12 cases noted to infect CNS[15]. Most of the cases of CNS infections were noted to be community acquired. The organism has been reported to cause infections in all age groups including children and neonates[5,20] however we noticed that all our patients reported with CNS infections were adults. It was intriguing to notice that most patients with CNS infections were immunocompetent, even though it was previously known to infect immunocompromised patients because of its poor virulence. In majority of S paucimobilis infections the prognosis is favourable however it has caused morbidity as well as mortality[2] in a limited number of instances. Most of the cases noted in our review had acute history and predominantly were adults with a few cases being senior citizens. The organism is easily identifiable using conventional and Vitek 2 method of isolation. There were few occurrences of coinfections, one patient had Listeria monocytogenes bacteraemia[10] and one had mycobacterium tuberculosis meningitis[7]. All the patients were treated mainly with antibiotics with an average duration of 21 days, ceftriaxone and meropenem were among the ones commonly administered, only two cases received additional steroids. Ventriculitis, hydrocephalus and IVH were the noted complications needing surgical interventions for EVD placement and VP shunting. Although they were immunocompetent and had no concomitant conditions, two individuals with hydrocephalus who had undergone EVD placement had a catastrophic outcome.

The antibiotic susceptibility pattern differed greatly throughout the case reports, indicating the importance of personalised decision making when treating CNS infections. Most of the isolates were sensitive to aminoglycosides, cephalosporins (3rd and 4th generation) and meropenem. Colistin was frequently encountered to be resistant.

The study undertaken by Bayram N, *et al* included a total of 24 children, one of which had CNS infection but lacked comprehensive clinical information. Similarly, another research with four instances of CNS infection lacked a clear description of the patient features, limiting the ability to analyse those cases[5,6].

Our review had few limitations, we attempted to include all literature relating to CNS infection caused by S paucimobilis, although several studies did not offer a full description of the patients. Our extremely small sample size made it difficult to generalise our results, which was another downside. The strength of this study is that we attempted to analyse the limited literature extensively to help disseminate the information to help aid in the treatment of subsequent cases. Our review would signify the importance of early recognition and treatment of a condition which can at times be fatal. Further prospective studies are needed to better understand the clinical implications of this organism.

Conclusion

Sphingomonas paucimobilis is an occasional human pathogen. The bacteria can cause both nosocomial and community-acquired infections. It has been implicated to affect humans irrespective of their immune status. S Paucimobilis can cause infections of various organs however there are only 13 cases of infections of CNS in the searched literature. Though most of the cases have favourable outcome but the organism can cause complications like hydrocephalus and intraventricular haemorrhage and can be fatal leading to death. The infection responds well to antibiotics thus emphasising on the need for early identification and initiation of appropriate antibiotics in suspected cases.

Conflict of interest : None of the authors have any conflicts of interest associated with the work presented in this manuscript.

Ethics approval and informed consent According to regional regulations, there was no need for ethical approval for this evaluation.

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Data availability All data generated or analyzed during this study are included in this article.

Further enquiries can be directed to the corresponding author.

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