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An uncommon encounter: *Pseudescherichia vulneris* infection in a neonate. A case report

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ABSTRACT:

— ***Background:*** *Pseudescherichia vulneris* is a rare pathogen associated with infections in immunocompromised adults. This may be the first documented report of a neonate with late-onset sepsis.

— ***Case Report:*** An 18-day-old term baby was admitted to the pediatric unit with a five-day history of fever and refusal to feed. The infant was irritable, jaundiced, and pale. Notably, the mother experienced prolonged premature rupture of membranes (PROM). Blood culture isolated multi-drug-resistant *P. vulneris* and *Acinetobacter baumannii*. Identifying the organism was challenging due to its rarity, phenotypic similarity to other Enterobacteriaceae, and limited access to advanced diagnostic tools. The treatment was successful, as the baby responded well to antibiotics and was discharged.

— ***Conclusions:*** This case highlights the potential for rare pathogens to cause neonatal sepsis, the challenges in diagnosing such infections in resource-limited settings, and the risk of multidrug-resistant strains emerging in the isolate. The case also underscores the necessity for further research to elucidate the pathogenesis, epidemiology, and optimal management of *P. vulneris* infections.

— ***Keywords:*** *Pseudescherichia vulneris, Infection, Neonate, Rare, Case report.*

BACKGROUND

Pseudescherichia vulneris is a Gram-negative, facultatively anaerobic, motile bacillus belonging to the family Enterobacteriaceae[1,2]. It exhibits a broad host range, colonizing humans, animals, and the environment[1]. Initially classified as Alma group 1 and Enteric group 1 based on phenotypic characteristics, this organism was subsequently reclassified as *Escherichia vulneris* following DNA hybridization studies[1]. However, advancements in genotypic and genomic identification techniques, particularly phylogenomic, revealed its distinct evolutionary lineage. This new understanding necessitated a further reclassification, resulting in the current designation of *Pseudoescherichia vulneris*[2,3] exemplifying the dynamic nature of bacterial taxonomy. It has been implicated in infections affecting both immunocompetent and immunocompromised individuals, manifesting as a sole pathogen or in polymicrobial infections. The spectrum of reported infections encompasses both localized presentations (e.g., wound infections, localized peritonitis, osteomyelitis, arthritis)[1,4-8] and systemic manifestations (bacteremia, urosepsis, gastroenteritis with sepsis, and meningitis)[9-12]. Notably, the majority of documented cases have involved adults and young

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children, with the youngest reported patient being a 2-month-old infant[9]. This case report may present the first documented instance of *P. vulneris* co-infection in a neonate, contributing valuable insights into its clinical presentation, diagnosis, and treatment in resource-limited settings.

CASE PRESENTATION

An 18-day-old female neonate, born at term *via* spontaneous vaginal delivery following a documented history of prolonged premature rupture of membranes (PROM), presented to the Special Care Baby Unit with a five-day history of poor feeding and jitteriness. She was delivered at a peripheral healthcare facility, and while the APGAR score was not communicated to the parents, the infant exhibited a vigorous cry at birth. Physical examination revealed an irritable neonate with moderate pallor and jaundice. Anthropometric measurements documented a normal weight of 2.65 kg, length of 48 cm, and head circumference of 35 cm. Based on clinical presentation, a diagnosis of suspected late-onset sepsis (LOS) was made. The neonate was commenced on intravenous ampicillin (50 mg/kg/dose every 6 hours) and gentamicin (2.5 mg/kg/dose every 12 hours) following the unit protocol for empirical treatment of LOS.

Complete blood count (CBC) revealed anemia with a packed cell volume (PCV) of 37% and hemoglobin of 12.3 g/dl. The white blood cell (WBC) count was within the normal range at 7.52 x 10^9/L. However, the differential count showed a lower border normal absolute neutrophil count (ANC) of 2,016/L, with an elevated percentage of immature neutrophils (band forms: 9%) and polymorphonuclear neutrophil (PMN) count of 677/L. Additionally, there was evidence of thrombocytosis with a platelet count of 529 x 10^9/L. The calculated I:M (immature to mature neutrophil) and I:T (immature to total neutrophil) ratios were elevated at 0.5 and 0.32, normal values are 0.2 and 0.3, respectively.

Inflammatory biomarkers were assessed. Procalcitonin (PCT) was elevated at 0.32 ng/mL (normal range: 0.25 ng/mL), while high-sensitivity C-reactive protein (hs-CRP) and C-reactive protein (CRP) remained within normal reference ranges of 0.25 mg/L and 5 mg/L, respectively.

Blood culture yielded a polymicrobial infection, with the growth of two distinct bacterial pathogens confirming the initial diagnosis of suspected sepsis: a Gram-negative, motile, lactose-fermenting rod and a non-motile, non-lactose-fermenting organism. Further biochemical tests using the Microbact 24E system (BioMérieux, Marcy-l'Étoile, France) identified the organisms as *P. vulneris* and *Acinetobacter baumannii* with a bit of challenge, confirming the initial diagnosis of LOS. No repeat blood or other cultures were taken. Antibiotic susceptibility testing was performed using the Modified Kirby-Bauer disc diffusion method according to Clinical and Laboratory Standards Institute (CLSI) 2019 guidelines. *Pseudescherichia vulneris* exhibited sensitivity only to gentamicin and meropenem, while *A. baumannii* displayed susceptibility to all antibiotics tested. Tables 1 and 2 below show the biochemical test result and antibiotic susceptibility pattern. The neonate responded well to treatment with resolution of presenting symptoms and improvement in feeding. The patient was discharged home after seven days.

Table 1. Biochemical tests' reaction pattern of *P. vulneris.*

Tests	*P. vulneris*
Oxidase	Negative
Indole	Negative
Urease	Negative
Citrate	Negative
Ornithine	Positive
Hydrogen sulphite	Positive
Voges-proskeuer	Positive
Tryptophan-deaminase	Positive
Inositol	Positive
Sucrose	Positive
Sorbitol	Positive
Lysine	Positive
Glucose	Positive
Mannitol	Positive
O-*nitrophenyl*-B-D-*galactopyronoside*	Positive
Malonate	Positive
Raffinose	Positive
Arabinose	Positive
Maltose	Positive
Rhamnose	Positive
Xylose	Positive
Lactose	Positive
Argentine	Positive

DISCUSSION

Pseudescherichia vulneris infections are uncommon in humans and typically considered opportunistic. Initial isolations were from polymicrobial wound infections[1]. Notably, *P. vulneris* has been implicated in both localized and systemic infections, acting as either a sole causative agent[5,6,8-13] or co-existing with other bacterial pathogens[1,4]. In our case, *P. vulneris*

Table 2. Antibiotic susceptibility pattern of *P. vulneris.*

Antibiotic	Susceptibility test
Gentamicin	Sensitive
Meropenem	Sensitive
Amoxycillin-clavulanate	Resistant
Ceftriaxone	Resistant
Ceftazidime	Resistant
Ciprofloxacin	Resistant

was co-isolated with pathogen *A. baumanii* during a septic episode. This finding contrasts with other reports where *P. vulneris* was the sole causative agent of sepsis in an infant and older children[7-9,11]. The presence of multiple pathogens in this case makes it difficult to definitively determine the specific role of *P. vulneris* in the development of the neonate's sepsis. Infections are typically reported in immunocompromised adults or elderly individuals[5,6,10,12,13]. This case deviates from this pattern, presenting in a neonate. While neonates are not typically classified as immunosuppressed, their immune systems are immature compared to adults. This relative immune incompetence may have contributed to the neonate's susceptibility to infection, even though documented cases of infection exist in immunocompetent children[7-9]. The precise route of acquisition of human infections frequently remains unclear. Documented cases have linked the organism to contaminated medical devices, such as urinary[10] or peritoneal catheters[13], and wooden foreign bodies[8]. However, the source of infection in many reported cases, including this one, remains elusive[4,5,9]. We can only hypothesize about potential transmission routes. One possibility is vertical transmission from the colonized maternal genitourinary tract during delivery. This is particularly plausible given the presence of prolonged PROM in the mother, which could facilitate bacterial ascent. Additionally, the documented isolation of *P. vulneris* from the genitourinary tract supports this hypothesis[1]. Alternatively, acquisition from contaminated water sources cannot be entirely excluded. Documented evidence exists for the presence of the pathogen in some potable water supplies[1,14]. While the neonate was primarily breastfed, supplemental water feedings were also administered, creating a potential route for exposure.

The neonate presented with non-specific clinical features indistinguishable from those associated with sepsis from other etiologies. While our case involved *P. vulneris* bacteraemia, which has been documented in some reports[12,13], it differed from other presentations of the infection, which are site-specific (gastroenteritis, peritonitis, and urosepsis).

Hematological evaluation in our neonate revealed a CBC consistent with sepsis despite a normal WBC count. Normal WBCs have been documented in some reports[4,11]. This differs from leucocytosis reported by Jain et al[9]. Other CBC parameters, including ANC, I:M, and I:T ratio, deviated from expected ranges, suggesting an inflammatory response. Notably, our patient presented with anemia. This was also documented by Mohanty et al[11], while Jain et al[9] documented a normal hematocrit level. Interestingly, both our investigation and the study by Jain et al[9] identified thrombocytosis, a finding suggestive of a reactive process. In contrast to the elevated CRP levels reported by Jain et al[9], our study found an elevated procalcitonin (PCT) level despite normal CRP. This finding stresses the superior sensitivity and specificity of PCT as a diagnostic marker for neonatal infections compared to CRP.

Our facility was fortunate to identify the organism using the traditional structural phenotype and biochemical testing methods despite limitations compared to advanced molecular characterization techniques like genomic sequencing and typing[2,3]. These techniques offer greater precision in organism identification but were not employed due to resource constraints.

This case highlights a significant issue: the scarcity of resources and expertise needed to identify the uncommon pathogen *P. vulneris*, a problem commonly encountered in resource-limited healthcare settings such as those found in many developing countries, including Nigeria. Consequently, there is a significant risk of misdiagnosis or missed diagnoses of such infections, potentially leading to inappropriate treatment and adverse outcomes. It also serves as a stark reminder of the critical need to implement strategies that will enhance diagnostic capabilities across all healthcare settings. Prioritizing such advancements can improve patient health outcomes significantly.

The organism is generally reported to exhibit susceptibility to a broad spectrum of antibiotics. However, in this case, the organism displayed resistance to multiple antimicrobials with susceptibility limited to gentamicin and meropenem, similar resistance profiles have been documented[11,15]. This observation deviates from the previously documented reported susceptibility profile[1,4,6,9,10,12,16]. The observed resistance pattern highlights the potential for the emergence of antibiotic resistance within this species and may probably be related to the prevalent poor antibiotic stewardship in our environment. This observation warrants further investigation and underscores the importance of continuous antimicrobial resistance surveillance programs for bacterial pathogens generally. The rarity and the challenges associated with identifying this organism, particularly in our facility encountering its first case, may have contributed to a delay in definitive antimicrobial susceptibility testing and, as a result, became available towards the time of discharge. Fortunately, initial empiric therapy with gentamicin, one of the effective antibiotics against this isolate, proved successful.

The prognosis for *P. vulneris* infection appears favorable, with most documented cases[4-6,9-11,13], including this one, reporting successful patient discharge. Few reports provide limited information on its outcome[1,8]. Data on long-term outcomes is limited.

CONCLUSIONS

We report a rare case of *P. vulneris* co-infection in a neonate, demonstrating successful clinical management. We notably achieved successful identification of this uncommon pathogen, even within the constraints of a resource-limited environment with limited access to advanced diagnostic tools. We also highlighted the possibility of emerging antimicrobial resistance in the pathogen.

Informed Consent:
Written informed consent was obtained from the caregiver of the patient to publish this case report.

Ethics Approval:
The design of the work was approved by the Health Research Ethics Committee of Ahmadu Bello University Teaching Hospital Zaria with ref. No. ABUTH/HREC/CL/05.

Availability of Data and Materials:
The data that support the findings are available on request from the corresponding author.

Conflict of Interest:
The authors affirm that they have no competing financial interests or personal relationships that could have influenced the work reported in this paper.

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Authors' Contributions:
SSM: conceived of the study, contributed to the literature search, reviewed the case, and drafted the manuscript.
AZM: conceived the study, did the literature search, summarized the case, and co-drafted the manuscript.
IM: organism isolation and identification, and reviewed the manuscript.
MSI: organism isolation/identification, antimicrobial susceptibility test, and reviewed the manuscript.
SA: organism identification, antimicrobial susceptibility test, and reviewed the manuscript.
IA: conceived the study, contributed to literature search, reviewed the case summary and overall review of the manuscript.

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The authors utilized [ChatGPT/Curie] during the preparation of this work to help clarify their thoughts and improve grammatical accuracy. Following the use of this tool/service, the authors reviewed and edited the content as necessary and assumed full responsibility for the final publication.

References

1. Brenner DJ, McWhorter AC, Knutson KL, Steigerwalt AG. Escherichia vulneris: A new species of Enterobacteriacea associated with human wounds. J Clin Microbiol 1982; 15: 1133-1140.
2. Alnajar S, Gupta RS. Infection, Genetics and Evolution Phylogenomics and comparative genomic studies delineate six main clades within the family Enterobacteriaceae and support the reclassi fi cation of several polyphyletic members of the family. Infect Genet Evol 2017; 54: 108-127.
3. Yu D, Banting G, Neumann NF. A review of the taxonomy, genetics, and biology of the genus escherichia and the type species escherichia coli. Can J Microbiol 2021; 67: 553-571.
4. Jepsen CF, Klebe TM, Prag J. Escherichia vulneris in a Danish soccer wound. Scand J Infect Dis 1997; 29: 313-314.
5. Añón MT, Ruiz-Velasco LM, Borrajo E, Giner C, Sendino M, Cantón R. Escherichia vulneris infection. Report of 2 cases. Enferm Infecc Microbiol Clin 1993; 11: 559-561.
6. Senanayake SN, Jadeer A, Talaulikar GS, Roy J. First reported case of dialysis-related peritonitis due to Escherichia vulneris. J Clin Microbiol 2006; 44: 4283-4284.
7. Pérez C, Melo P, Besomi J, Porte L, Wilhelm P. Escherichia vulneris. Caso Clin 2018; 35: 80-82.
8. Levine WN, Goldberg MJ. Escherichia vulneris osteomyelitis of the tibia caused by a wooden foreign body. Orthop Rev 1994; 23: 262-265.
9. Jain S, Nagarjuna D, Gaind R, Chopra S, Debata PK, Dawar R, Sardana R, Yadav M. Escherichia vulneris: an unusual cause of complicated diarrhoea and sepsis in an infant. A case report and review of literature. New Microbes New Infect 2016; 13: 83-86.
10. Awsare SV, Lillo M. A case report of Escherichia vulneris urosepsis. Rev Infect Dis 1991; 13: 1247-1248.
11. Mohanty S, Chandra SP, Dhawan B, Kapil A, Das BK. Meningitis due to Escherichia vulneris. Neurol India 2005; 53: 122-123.
12. Kilani B, Ammari L, Chaabane HTBT Ben, Fendri C. Escherichia vulneris as a cause of bacteremia in a patient with chronic lymphocytic leukemia. Int J Infect Dis 2008; 12: 108-110.
13. Anne SC, Rothman AL. Escherichia Vulneris as a Cause of Intravenous Catheter-Related Bacteremia. Clin Infect Dis 2011; 44: 51-66.
14. Querler LL, Donnio PY, Poisson M, Rouzet-Gras S, Avril JL. Isolation of Escherichia vulneris in drinking water. Ann Biol Clin (Paris) 1997; 55: 33-35.
15. Chaudhury A, Nath G, Tikoo A, Sanyal SC. Enteropathogenicity and Antimicrobial Susceptibility of New Escherichia Spp. J Diarrhoeal Dis Res 1999; 17: 85-87.
16. Stock I, Wiedemann B. Natural antibiotic susceptibility of Escherichia coli, Shigella, E. vulneris, and E. hermannii strains. Diagn Microbiol Infect Dis 1999; 33: 187-199.