

World Journal of *Clinical Cases*

World J Clin Cases 2023 August 6; 11(22): 5193-5415



MINIREVIEWS

- 5193 Research progress on reactive oxygen species production mechanisms in tumor sonodynamic therapy
Dong HQ, Fu XF, Wang MY, Zhu J

ORIGINAL ARTICLE**Retrospective Study**

- 5204 Combining the age-male-albumin-bilirubin-platelets score and shear wave elastography stratifies carcinogenic risk in hepatitis C patients after viral clearance
Masaoka R, Gytoku Y, Shirahashi R, Suda T, Tamano M
- 5215 Changes in neurotransmitter levels, brain structural characteristics, and their correlation with PANSS scores in patients with first-episode schizophrenia
Xu XJ, Liu TL, He L, Pu B
- 5224 Five-year outcomes of immediate implant placement for mandibular molars with and without chronic apical periodontitis: A retrospective study
Yang H, Luo D, Yuan MJ, Yang JJ, Wang DS

Observational Study

- 5236 Standardization of apple cancellation test for neglect patients in Korea: An observational study
Jang WH, Jang JS

Prospective Study

- 5244 Diabetic neuropathy results in vasomotor dysfunction of medium sized peripheral arteries
Ege F, Kazci Ö, Aydin S

SYSTEMATIC REVIEWS

- 5252 COVID-19-induced gastrointestinal autonomic dysfunction: A systematic review
Elbeltagi R, Al-Beltagi M, Saeed NK, Bediwy AS

META-ANALYSIS

- 5273 Meta-analysis of outcomes from drug-eluting stent implantation in infrapopliteal arteries
Li MX, Tu HX, Yin MC

CASE REPORT

- 5288 Acute hepatitis of unknown etiology in an adult female: A case report
Dass L, Pacia AMM, Hamidi M

- 5296** Zimberelimab plus chemotherapy as the first-line treatment of malignant peritoneal mesothelioma: A case report and review of literature
Peng XD, You ZY, He LX, Deng Q
- 5303** Recurrent ventricular arrhythmia due to aconite intoxication successfully treated with landiolol: A case report
Matsuo C, Yamamoto K, Fukushima H, Yajima D, Inoue H
- 5309** Anti-phospholipase A2 receptor-associated membranous nephropathy with human immunodeficiency virus infection treated with telitacicept: A case report
Wang JL, Sun YL, Kang Z, Zhang SK, Yu CX, Zhang W, Xie H, Lin HL
- 5316** Rapid progression of heart failure secondary to radioactive iodine treatment of hyperthyroidism: A case report
Li ZH, Ni LJ, Liu YQ, Si DY
- 5322** Pathological complete response to neoadjuvant alectinib in unresectable anaplastic lymphoma kinase positive non-small cell lung cancer: A case report
Wang LM, Zhao P, Sun XQ, Yan F, Guo Q
- 5329** Hepatoid adenocarcinoma of the stomach with neuroendocrine differentiation: A case report and review of literature
Fei H, Li ZF, Chen YT, Zhao DB
- 5338** Acquired haemophilia as a complicating factor in treatment of non-muscle invasive bladder cancer: A case report
Ryšánková K, Gumulec J, Grepl M, Krhut J
- 5344** Persistent dysexecutive syndrome after pneumococcal meningitis complicated by recurrent ischemic strokes: A case report
Abbruzzese L, Martinelli G, Salti G, Basagni B, Damora A, Scarselli C, Peppoloni G, Podgorska A, Rosso G, Bacci M, Alfano AR, MANCUSO M
- 5351** Treatment of refractory anti-melanoma differentiation-associated gene 5 antibody-positive dermatomyositis complicated by rapidly progressing interstitial pulmonary disease: Two case reports
Wang QH, Chen LH
- 5358** TINAVI robot-assisted one-stage anteroposterior surgery in lateral position for severe thoracolumbar fracture dislocation: A case report
Ye S, Chen YZ, Zhong LJ, Yu CZ, Zhang HK, Hong Y
- 5365** Individual with concurrent chest wall tuberculosis and triple-negative essential thrombocythemia: A case report
Xu XY, Yang YB, Yuan J, Zhang XX, Kang L, Ma XS, Yang J
- 5373** Self-strangulation induced penile partial amputation: A case report
Maimaitiming ABLT, Mulati YLSD, Apizi ART, Li XD
- 5382** Long-term rare giant sialolithiasis for 30 years: A case report and review of literature
Mao JS, Lee YC, Chi JCY, Yi WL, Tsou YA, Lin CD, Tai CJ, Shih LC

- 5391** Kawasaki disease with peritonsillar abscess as the first symptom: A case report
Huo LM, Li LM, Peng HY, Wang LJ, Feng ZY
- 5398** Treatment of a patient with severe lactic acidosis and multiple organ failure due to mitochondrial myopathy: A case report
Chen L, Shuai TK, Gao YW, Li M, Fang PZ, Christian W, Liu LP
- 5407** Early esophageal carcinomas in achalasia patient after endoscopic submucosal dissection combined with peroral endoscopic myotomy: A case report
An BQ, Wang CX, Zhang HY, Fu JD

LETTER TO THE EDITOR

- 5412** Caution in the use of sedation and endomyocardial biopsy for the management of pediatric acute heart failure caused by endocardial fibroelastosis
Xin XX, Se YY

ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Etienne Andrade Munhoz, PhD, Associate Professor, Department of Dentistry, Health Science Centre, Federal University of Santa Catarina, Florianopolis 88040-379, Brazil. etiamfob@yahoo.com

AIMS AND SCOPE

The primary aim of *World Journal of Clinical Cases* (WJCC, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

INDEXING/ABSTRACTING

The WJCC is now abstracted and indexed in Science Citation Index Expanded (SCIE, also known as SciSearch®), Journal Citation Reports/Science Edition, Current Contents®/Clinical Medicine, PubMed, PubMed Central, Reference Citation Analysis, China National Knowledge Infrastructure, China Science and Technology Journal Database, and Superstar Journals Database. The 2023 Edition of Journal Citation Reports® cites the 2022 impact factor (IF) for WJCC as 1.1; IF without journal self cites: 1.1; 5-year IF: 1.3; Journal Citation Indicator: 0.26; Ranking: 133 among 167 journals in medicine, general and internal; and Quartile category: Q4.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Si Zhao; Production Department Director: Xu Guo; Editorial Office Director: Jin-Lei Wang.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Bao-Gan Peng, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati, Ja Hyeon Ku

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

August 6, 2023

COPYRIGHT

© 2023 Baishideng Publishing Group Inc

INSTRUCTIONS TO AUTHORS

<https://www.wjgnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjgnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>



Persistent dysexecutive syndrome after pneumococcal meningitis complicated by recurrent ischemic strokes: A case report

Laura Abbruzzese, Giulia Martinelli, Giulia Salti, Benedetta Basagni, Alessio Damora, Cristiano Scarselli, Giulia Peppoloni, Aleksandra Podgorska, Giuliana Rosso, Marco Bacci, Alba Rosa Alfano, MAURO MANCUSO

Specialty type: Medicine, research and experimental

Provenance and peer review:

Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0

Grade B (Very good): 0

Grade C (Good): C, C, C

Grade D (Fair): 0

Grade E (Poor): 0

P-Reviewer: Arboix A, Spain; Tang ZP, China

Received: April 24, 2023

Peer-review started: April 24, 2023

First decision: May 31, 2023

Revised: June 9, 2023

Accepted: July 17, 2023

Article in press: July 17, 2023

Published online: August 6, 2023



Laura Abbruzzese, Giulia Martinelli, Giulia Salti, Benedetta Basagni, Alessio Damora, Cristiano Scarselli, Giulia Peppoloni, CRT, Tuscany Rehabilitation Clinic, Montevarchi, Arezzo 52025, Italy

Aleksandra Podgorska, Giuliana Rosso, Marco Bacci, MAURO MANCUSO, Physical and Rehabilitative Medicine Unit, NHS ASL-Tuscany South Est, Grosseto 58100, Italy

Alba Rosa Alfano, Department of Internal Medicine and Medical Specialties, UOC Geriatrics, Sapienza University of Rome, Rome 00185, Italy

Corresponding author: Benedetta Basagni, PsyD, CRT, Tuscany Rehabilitation Clinic, No. 1 Piazza Del Volontariato, Montevarchi, Arezzo 52025, Italy. bbasagni@gmail.com

Abstract

BACKGROUND

Meningitis is a possible complication of pneumococcal infection concerning acute otitis media and sinusitis. It might compromise cognitive function, both for the infection itself and the vascular events that sometimes follow the acute phase.

CASE SUMMARY

Here we describe the case of a 32-year-old female patient admitted to the emergency room due to extensive pneumococcal meningitis as a consequence of sinus outbreak. She presented with extensive laminar ischemic damage in the acute phase, resulting in severe cognitive and behavioural impairment. Four years of follow-up, through neuropsychological assessments and neuroradiological investigations, demonstrated the presence of subsequent vascular events, 3 months and 2 years after onset.

CONCLUSION

The case is discussed in light of scientific knowledge of the long-term outcomes of this pathology in order to potentially improve diagnosis and promote better outcomes.

Key Words: Pneumococcal meningitis; Stroke, Cognitive and behavioural functioning; Long-term outcome; Case report

©The Author(s) 2023. Published by Baishideng Publishing Group Inc. All rights reserved.

Core Tip: Meningitis is a possible complication of pneumococcal infection concerning acute otitis media and sinusitis. We describe the case of a 32-year-old female patient with extensive pneumococcal meningitis as a consequence of sinus outbreak. She presented with extensive laminar ischemic damage in the acute phase, resulting in severe cognitive and behavioural impairment. Four years of follow-up, through neuropsychological assessments and neuroradiological investigations, demonstrated the presence of subsequent vascular events, 3 months and 2 years after the onset. The global functional outcome was unfavorable.

Citation: Abbruzzese L, Martinelli G, Salti G, Basagni B, Damora A, Scarselli C, Peppoloni G, Podgorska A, Rosso G, Bacci M, Alfano AR, MANCUSO M. Persistent dysexecutive syndrome after pneumococcal meningitis complicated by recurrent ischemic strokes: A case report. *World J Clin Cases* 2023; 11(22): 5344-5350

URL: <https://www.wjgnet.com/2307-8960/full/v11/i22/5344.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v11.i22.5344>

INTRODUCTION

According to the National Health Surveillance Institute, the incidence of acute community bacterial meningitis (BM) was, in 2006, 23 per 100000 inhabitants, adding together all ages and all types of bacteria. In adults over the age of 24, the bacteria involved are: *Pneumococcus*, *meningococcus*, and, more rarely, *listeria mono cytogenes*, *haemophilus influenzae* and group B *streptococcus*[1]. In developing countries, the annual incidence of pneumococcal meningitis ranges from 1-2 to 20/100000. Sites of infection may concern acute otitis media, mastoiditis, sinusitis, pneumonia, and endocarditis. Clinical manifestations include sudden high fever, headache, nausea, vomiting, confusion, drowsiness, convulsion, and in more severe cases, loss of consciousness. Mortality is about 20%.

Long-term studies on survivors have demonstrated that adults after pneumococcal meningitis are at high risk of neurological and neuropsychological deficits impairing daily life activities and quality of life. In the Kloek *et al*[2] study, a cohort of 79 patients was evaluated 1 to 5 years after acute illness. After a median of 2 years, 34% of the sample had persistent neurologic sequelae, most commonly hearing loss (27%). On overall neuropsychological evaluation, patients performed worse than the controls. Primary cognitive deficits as cognitive slowness, associated with longer reaction times, were described[3,4]. In addition, alterations in executive function have also been reported, with prevalent working memory deficits, difficulty in inhibitory control of automatic responses, visuospatial reasoning, non-verbal learning, and verbal abstraction[3-6]. The alternating subcomponent of attention may also be compromised, together with impairments in the visual-constructive domain and in the amnesic visuospatial domain[3,4,6].

Neurological complications are frequent after pneumococcal meningitis[7]. Arterial stroke occurs in up to 30% of patients, cerebral venous thrombosis in up to 9%, and intracerebral hemorrhage in up to 9%[8]. In the study by Schmidt *et al*[5], brain swelling was observed in 24% of patients, small vessel vasculitis in 8.5%, impairment of cerebrospinal fluid circulation in 7%, and sinus thrombosis in 3%.

Cerebral infarction typically develops within the first few days of the disease when the central nervous system inflammation is most severe. Indeed, meningeal inflammation can lead to profound vascular alterations on the meningeal vessels, creating a vasculitis that participates in cerebral anoxia and alterations in blood flow. Delayed cerebral vasculopathy is a rare, but possible complication[9].

We hereby report a case of a young woman affected by severe and persistent cognitive sequelae after pneumococcal meningitis due to extensive sinusitis, who presented repeated ischemic damages. We describe the clinical history, the cognitive-behavioural profile, and the functional outcome, during the four years following onset. Written informed consent was obtained from the patient for publication of this case report.

CASE PRESENTATION

Chief complaints

A 32-year-old female presented to the emergency department complaining hyperpyrexia, headache, and neck pain.

History of present illness

The patients' symptoms started a few days earlier and had worsened in the last 12 h. The patient was pregnant at 39 + 1 wk of gestation and underwent emergency cesarean delivery due to the severity of the clinical condition.

History of past illness

The patient was completely independent in everyday activities and she had no previous neurological disorders.

Personal and family history

The patient was a right-handed married female educated to 16 years of age, an artisan by profession. The patient had a three-year-old son born from a eutocic delivery and was pregnant with a second child upon admission to the hospital.

Physical examination

Physical examination showed symptoms consistent with a clinical pattern of meningitis.

Laboratory examinations

The lumbar puncture was positive for meningitis. The patient also underwent hematological tests to assess thrombotic risk and was assessed with computed tomography-angiography of the cerebral circulation to exclude potential atherosclerotic plaques. In order to exclude potential artery/venous shunts, a transcranial doppler with bubble test was conducted. The results showed the absence of potential causes for stroke. Additional hematologic exams to exclude autoimmunity pathologies were conducted and were negative.

Imaging examinations

The brain magnetic resonance imaging (MRI) performed revealed bilateral T2 hyperintensity of the cortex signal in large brain areas. The fronto-polar, insular, fronto-medial and cingulate bilateral regions were affected, with a prevalence of damage on the right side. On this side, the frontal operculum and the inferior lobule, and, to a lesser extent, the superior parietal one, were more clearly involved. The neuroimaging was compatible with extensive laminar ischemic damage in the aforementioned areas.

FINAL DIAGNOSIS

The final diagnosis of the presented case is pneumococcal meningitis, with sinus outbreak.

TREATMENT

The patient underwent pharmacological treatment with antibiotics and cortisone.

OUTCOME AND FOLLOW-UP

One month after onset, the patient was transferred to a neurorehabilitation unit for motor and cognitive rehabilitation. The results of the assessments carried out, both during this period of hospitalization and after discharge, are described below. [Figure 1](#) shows the timeline of the main stages of the rehabilitative training, as well as the different neurological and neuropsychological assessments and neuroradiological examinations, conducted over the range of 4 years. The results of each neuropsychological test performed over time are presented in [Table 1](#). Some tests were found to be inexecutable, due to the lack of attention skills.

The neurological examination (T0) performed upon admission to the rehabilitation ward showed the presence of isochoric pupils, isocyclic pupils, reactive to direct and consensual light stimulus as well as present pharyngeal reflex, trigeminal reflexes, coughing and vomiting. The patient showed symmetrical tendon reflexes, plantar reflex in bilateral flexion, non-elicitable Hoffmann, and bilaterally mentioned prehension reflex. The motor skills were preserved, with the execution of even complex activities, even if there were also automatic and aimless movements. The patient was not self-sufficient and required total assistance in most daily activities [functional independence measure (FIM)[\[10\]](#), FIM total: 25, motor FIM: 18, cognitive FIM: 7].

The patient showed herself to be alert, but high distractibility was present as irrelevant stimuli caught her attention. She visually engaged the examiner in all spatial positions and showed occasional imitative responses towards significant others in a framework of inertia, apathy, and the absence of motor and verbal initiative. Due to insufficient attentional resources, the neuropsychological evaluation was not yet administrable in this initial phase.

Three months after the onset and during the rehabilitation process, due to the sudden worsening of neurological conditions, the patient underwent a new MRI which showed the known hyperintensity of the signal in fluid attenuated inversion recovery (FLAIR) at the level of the cortex bilaterally. In particular, the temporal-mesial, insular, frontal-opercular and frontal areas were affected, with a slight right prevalence present. A new moderate cortical hyperintensity was found at the level of the right pre and post-rolandic cortex area and in the region of the ipsilateral angular gyrus. MRI also showed a net enlargement of the ventricular system, specifically the frontal and temporal horns, and of the periencephalic liquor sulci. In particular, the Sylvian fissures were enlarged bilaterally, due to pronounced atrophic outcomes (medial temporal lobe atrophy, score 3-4). Additionally, the corpus callosum was thinned.

Due to a significant behavioural change, a neuropsychological assessment was performed (T1). The profile was characterized by decontrolled behaviour, with distractibility, inability to inhibit irrelevant stimuli concerning environmental demands, disinhibition, hyperactivity leading to impulsive agitation, and automatic utilization behaviours. In addition, we observed temporal, spatial, and personal disorientation. Anosognosia was also observed. Communicative abilities began to convey essential needs, interspersed with automatic perseverative expressions, such as "Can we go outside?", "Is my husband there?", "Can you give me some food?". Neuropsychological tests showed a severe dysexecutive syndrome with difficulty in controlling automatic responses and marked disturbance of attentional function, particularly for the selective and alternating subcomponents. The sustained attention was partially preserved. In the praxis-

Table 1 Timeline of behavioural disturbances.

Cluster	Behavioural impairment	T ₀	T ₁	T ₂	T ₃	T ₄
Motivation disorders	Apathy					
	Reduced finalistic cognitiveness	+	+	+	+	+
	Reduced emotional reactivity	+	+	+	+	+
	Acinetic mutism	+	-	-	-	-
Disinhibition disorders	Impulsiveness					
	Motor	+	+	+	+	+
	Cognitive	+	+	+	+	+
	Social/personal behaviour impairments					
	Bizarre/inadequate behaviour	+	+	+	+	+
	Invasive behaviour	-	+	+	-	-
	Hyperorality	-	+	+	-	-
	Hyperactivity	-	+	+	-	-
	Afinalistic movements	+	+	+	+	+
	Dromomania	-	-	+	+	+
	Negligence in personal care	+	+	+	+	+
	Environmental dependency	+	+	+	-	-
	Aggressiveness	-	-	-	-	-
	Acquired sociopathy					
	Difficulty in recognising emotional expressions	-	-	-	-	-
	Difficulty in inferring other people's emotions	-	-	-	-	-
	Reduced theory of mind	+	+	+	+	+
Perseverative disorders	Motor	+	+	+	+	+
	Cognitive	+	+	+	+	+
Anosognosia		+	+	-	+	+

constructive domain, the performance qualified as pathological due to difficulties in the executive planning of the realization of the graphic elements.

Six months after the onset (T2), spatial and personal orientation was recovered. The awareness was classifiable in the cognitive level of Crossons' model[11]: The patient verbally reported the neurological event that occurred as a result of the continuous exposure to that information during the rehabilitation training. Using a structured assessment, improvements in selective attention skills were observed. However, attention-shifting skills were still deficient. The dysexecutive syndrome, both for cognitive and behavioural aspects, remained severe. In addition, alterations in social conduct, invasive behaviours, hyperorality, and a tendency to dromomania were present. In the memory domain, the patient presented a main working memory deficit which, with a reduction in attention control skills, resulted in a limitation of storage and recall processes, mainly for verbal material. The dysexecutive profile affected the correct execution of the tests, leading to fluctuating performance.

Seven months after the onset, the patient was discharged for rehabilitation at the residence area. Upon returning home, the patient was able to carry out the activities of daily life only with the need of supervision or adaptations (total FIM: 98, motor FIM: 79, cognitive FIM: 19). From the motor point of view, she had regained adequate movement strength. Daily life management was significantly affected by the stability of the behavioural profile, which considerably compromised affective and social relationships.

The neuropsychological evaluation, performed at the follow-up 9 months after the neurological event (T3), showed an increase in accuracy, selection of attentional target, and attentional shifting skills. However, there was a regression of awareness aspects leading to anosognosia. The dysexecutive aspects and the memory difficulties previously noted were substantially stable. However, the loss of inhibitory control began to affect the sexual sphere, resulting in significant behavioural management difficulties. In the praxis-constructive domain, although a greater number of elements have been copied, a pathological performance persisted due to difficulties in the executive planning of the realization of the

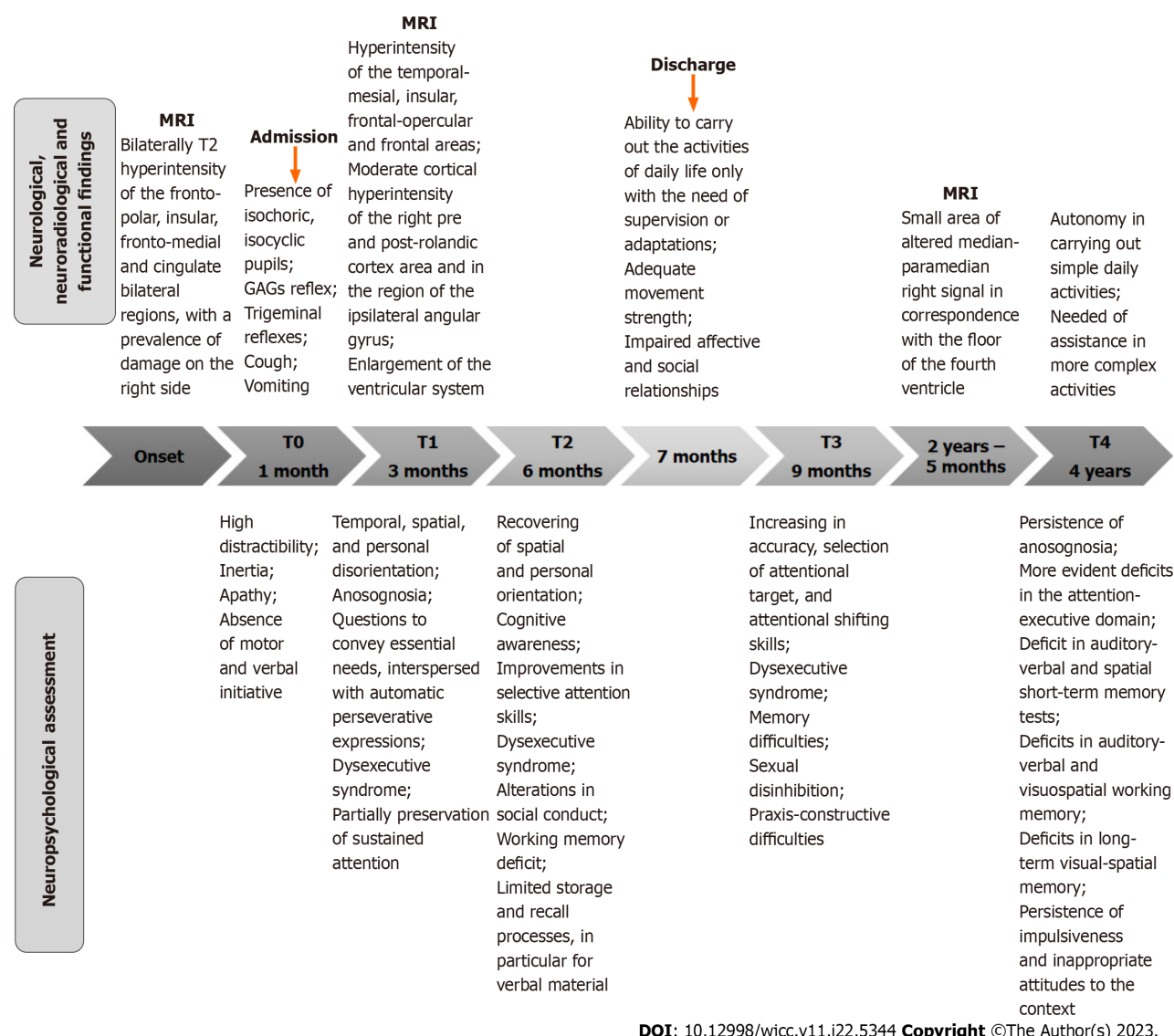


Figure 1 Case report timeline. MRI: Magnetic resonance imaging.

graphic elements.

At that time, the patient followed attending a recreational center, for people with disabilities where she undertook cognitive stimulation training, with ecological objectives aimed at recovering autonomy. With respect to her functional profile, she never went back to work and the parental role was delegated to her husband.

Two years and five months from onset, the patient suffered a new ischemic event in the pontine region. The MRI showed a small area of altered median-paramedian right signal in correspondence with the floor of the fourth ventricle, hyperintensity in the FLAIR, and long TR images, characterized by signs of restrictive edema in diffusion-weighted imaging and by nuanced impregnation after administration of contrast medium. The result was compatible with a recent pontine ischemic lesion. Apart from this new lesion, the remaining findings were compatible with the expected evolution of the previous brain damage.

At that time the patient started an antithrombotic therapy. Four years and two months after the onset, the patient underwent a new neuropsychological assessment (T4), which showed a substantially stable cognitive profile compared to the previous evaluation which was characterized by multi-domain difficulties. In particular, deficits in the attention-executive domain were more evident (distractibility due to interference from irrelevant stimuli, slowdown in information processing, deficit of attentional switching capacity, sensitivity to interference, and loss of inhibitory control and of cognitive flexibility). Regarding the memory system, scores at the lower limit of the norm were observed in both auditory-verbal and spatial short-term memory tests, as well as deficits in auditory-verbal and visuospatial working memory and in long-term visual-spatial memory. Relative to behaviour, compared to the previous evaluation, a slight improvement was observed (a decrease in sexual disinhibition and in-use behaviour, and the absence of hyperphagia). Instead, impulsiveness and sometimes inappropriate attitudes to the context remained. The patient was completely autonomous in carrying out simple daily activities but needed assistance with regard to more complex activities (Activities of Daily Living[12]: 6/6; Instrumental Activities of Daily Living[13]: 2/8). The patient was still anosognosic.

Regarding the rehabilitative interventions, the severity of the cognitive-behavioural framework had not always allowed the setting of structured training. In the initial phase, cognitive rehabilitation sessions focused on attentional exercises aimed at inhibiting the interference of irrelevant information and increasing the ability of sustained attention. Then, with the increasing of attentive skills, the rehabilitation focused on the reorganization of awareness and behaviour through cognitive-behavioural methods aimed at reorienting and monitoring impulsivity and verbal perseverations. The patient had also undergone selective and alternating attentional training.

Over time, the objectives have shifted from the restorative plan to the compensatory and functional efficiency plan, up to the inclusion in the recreation center.

DISCUSSION

Here we have described the case of a woman with severe cognitive sequelae, after BM starting from sinusitis and, consistent with the anatomical contiguity, prevalent damage in the frontal lobe, and consequently in executive functions and behaviour control. The patient presented dysexecutive syndrome with a severe behavioural disturbance. The profile has always been highly variable over time whereas it still remains serious. Symptoms are only partially alleviated, and significantly affect the resumption of independent life. Due to the cognitive and behavioural profile, the clinical observation was more informative than the neuropsychological assessment. Indeed, neuropsychological tests were greatly affected by attention impairment, compromising their administration and reliability. Overall, the patient mostly failed in all attentional and executive function tasks.

With respect to behavioural profile, in the post-acute phase, a Kluver-Bucy syndrome was even supposed. The syndrome follows bilateral lesions of the limbic system and manifests with personality alterations, hyperorality, hyperphagia, exhibitionism, compulsive hypersexuality, affective indifference, and aggression[14]. The patient presented with almost all the symptoms, except for aggression.

Compared to behavioural disorders of frontal origin often described in the literature[15], the patient presented a broad range of symptoms: Motivation disorders (reduced purposeful cognition, reduced emotional reactivity, and, in the initial phase, akinetic mutism), disinhibition disorders (motor and cognitive impulsivity, alterations in social conduct such as bizarre attitudes, sexual disinhibition, personhood, hyperactivity, dromomania, environmental dependency), perseverative behaviour and anosognosia.

After the severe first neurological damage, the patient had two more strokes in the following months and years which complicated the outcome. Stroke in young patients may be the manifestation of a specific haematological disease or may appear as a complication in the course of haematological disorders[16].

Nevertheless, thanks to the tests we conducted we tend to exclude this possibility, favoring the hypothesis of complications related to the infection. Indeed, it's known that infections affecting the central nervous system represent a rare cause of cerebral ischaemia. In the retrospective study of Arboix *et al*[17] of the 70 patients with cerebral ischemia from an unusual cause, 11 (15%) presented with central nervous system infections, such as syphilitic meningitis, infective endocarditis, meningococcal meningitis, pneumococcal meningitis, and human immunodeficiency virus infection. In our patient, the presence of vascular events repeated over time demonstrates how the suffering of the cerebral vessels on an inflammatory basis can involve a greater risk of occurring in new ischemic events, even at a distance from the acute phase.

The prognosis of our patient appears to have been particularly negative due to the sum of various factors. As already reported in the literature, BM, even in the absence of vascular complications, can lead to long-term alterations in cognitive functions[2-5]. A recent study hypothesized that macrophage migration inhibitory factor (MIF) upregulation, a proinflammatory cytokine, and a neuro-endocrine mediator, contributes to the cognitive impairments of survivors of pneumococcal meningitis[18]. Several studies showed the association between increased MIF production and Alzheimer disease suggesting MIF involvement in the neuro-inflammatory process occurring in mild cognitive impairment and in cognitive decline[19].

The case we reported, probably sums up the effects of primary inflammatory damage with those of secondary, repeated, vascular damage. Furthermore, despite undergoing many intensive cognitive rehabilitation training sessions, the improvements obtained over time were reduced. It is likely that the severe deficit of behavioural control has compromised the possibility of benefiting from rehabilitation.

Our case report presents some limitations. The lack of a neuropsychological battery sensitive to the severity of the patient's profile and administered at regular time intervals may have influenced the case description.

In summary, this case report shows a negative functional outcome in a young patient affected by BM, due to a severe cognitive and behavioural disturbance.

CONCLUSION

Understanding and evaluating cognitive profile in BM is a critical component of diagnosis and rehabilitative treatment, as it can cause severe complications and a poor prognosis for the patient. The description of this case report would highlight some of the clinical and neuropsychological features of BM with cerebrovascular complications to improve diagnosis and potentially promote better outcomes.

FOOTNOTES

Author contributions: Martinelli G, Salti G, Podgorska A, Rosso G, Bacci M, Scarselli C, Peppoloni G, contributed to manuscript data collection; Abbruzzese L and Basagni B writing and editing; MANCUSO M, Damora A, Alfano AR contributed to conceptualization and supervision; All authors have read and approved the final manuscript.

Informed consent statement: Informed written consent was obtained from the patient and her parents for the publication of this report and any accompanying images.

Conflict-of-interest statement: All the authors report no relevant conflicts of interest for this article.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution NonCommercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <https://creativecommons.org/licenses/by-nc/4.0/>

Country/Territory of origin: Italy

ORCID number: Benedetta Basagni 0000-0002-6173-584X.

S-Editor: Qu XL

L-Editor: A

P-Editor: Ju JL

REFERENCES

- 1 Stahl JP. Meningiti acute. *EMC-Neurologia* 2013; **13**: 1-11 [DOI: [10.1016/S1634-7072\(13\)64518-3](https://doi.org/10.1016/S1634-7072(13)64518-3)]
- 2 Kloek AT, Brouwer MC, Schmand B, Tanck MWT, van de Beek D. Long-term neurologic and cognitive outcome and quality of life in adults after pneumococcal meningitis. *Clin Microbiol Infect* 2020; **26**: 1361-1367 [PMID: [32006689](https://pubmed.ncbi.nlm.nih.gov/32006689/) DOI: [10.1016/j.cmi.2020.01.020](https://doi.org/10.1016/j.cmi.2020.01.020)]
- 3 van de Beek D, Schmand B, de Gans J, Weisfelt M, Vaessen H, Dankert J, Vermeulen M. Cognitive impairment in adults with good recovery after bacterial meningitis. *J Infect Dis* 2002; **186**: 1047-1052 [PMID: [12232850](https://pubmed.ncbi.nlm.nih.gov/12232850/) DOI: [10.1086/344229](https://doi.org/10.1086/344229)]
- 4 Hoogman M, van de Beek D, Weisfelt M, de Gans J, Schmand B. Cognitive outcome in adults after bacterial meningitis. *J Neurol Neurosurg Psychiatry* 2007; **78**: 1092-1096 [PMID: [17353256](https://pubmed.ncbi.nlm.nih.gov/17353256/) DOI: [10.1136/jnnp.2006.110023](https://doi.org/10.1136/jnnp.2006.110023)]
- 5 Schmidt H, Heimann B, Djukic M, Mazurek C, Fels C, Wallesch CW, Nau R. Neuropsychological sequelae of bacterial and viral meningitis. *Brain* 2006; **129**: 333-345 [PMID: [16364957](https://pubmed.ncbi.nlm.nih.gov/16364957/) DOI: [10.1093/brain/awh711](https://doi.org/10.1093/brain/awh711)]
- 6 Merkelbach S, Sittiger H, Schweizer I, Müller M. Cognitive outcome after bacterial meningitis. *Acta Neurol Scand* 2000; **102**: 118-123 [PMID: [10949529](https://pubmed.ncbi.nlm.nih.gov/10949529/) DOI: [10.1034/j.1600-0404.2000.102002118.x](https://doi.org/10.1034/j.1600-0404.2000.102002118.x)]
- 7 Kastenbauer S, Pfister HW. Pneumococcal meningitis in adults: spectrum of complications and prognostic factors in a series of 87 cases. *Brain* 2003; **126**: 1015-1025 [PMID: [12690042](https://pubmed.ncbi.nlm.nih.gov/12690042/) DOI: [10.1093/brain/awg113](https://doi.org/10.1093/brain/awg113)]
- 8 van de Beek D, Weisfelt M, de Gans J, Tunkel AR, Wijdicks EF. Drug Insight: adjunctive therapies in adults with bacterial meningitis. *Nat Clin Pract Neurol* 2006; **2**: 504-516 [PMID: [16932615](https://pubmed.ncbi.nlm.nih.gov/16932615/) DOI: [10.1038/ncpneu0265](https://doi.org/10.1038/ncpneu0265)]
- 9 Kato Y, Takeda H, Dembo T, Tanahashi N. Delayed recurrent ischemic stroke after initial good recovery from pneumococcal meningitis. *Intern Med* 2012; **51**: 647-650 [PMID: [22449677](https://pubmed.ncbi.nlm.nih.gov/22449677/) DOI: [10.2169/internalmedicine.51.6315](https://doi.org/10.2169/internalmedicine.51.6315)]
- 10 Hamilton BB, Laughlin JA, Fiedler RC, Granger CV. Interrater reliability of the 7-level functional independence measure (FIM). *Scand J Rehabil Med* 1994; **26**: 115-119 [PMID: [7801060](https://pubmed.ncbi.nlm.nih.gov/7801060/)]
- 11 Crosson B, Barco PP, Vellozo CA, Bolesta MM, Cooper PV, Werts D, Brobeck TC. Awareness and compensation in postacute head injury rehabilitation. *J Head Trauma Rehabil* 1989; **4**: 46-54
- 12 Meghan MG. Activities of daily living evaluation. ed. Encyclopedia of Nursing & Allied Health. Kristine Krapp. Gale Group, 2002
- 13 Cromwell DA, Eagar K, Poulos RG. The performance of instrumental activities of daily living scale in screening for cognitive impairment in elderly community residents. *J Clin Epidemiol* 2003; **56**: 131-137 [PMID: [12654407](https://pubmed.ncbi.nlm.nih.gov/12654407/) DOI: [10.1016/s0895-4356\(02\)00599-1](https://doi.org/10.1016/s0895-4356(02)00599-1)]
- 14 Lanska DJ. The Klüver-Bucy Syndrome. *Front Neurol Neurosci* 2018; **41**: 77-89 [PMID: [29145186](https://pubmed.ncbi.nlm.nih.gov/29145186/) DOI: [10.1159/000475721](https://doi.org/10.1159/000475721)]
- 15 Reber J, Tranel D. Frontal lobe syndromes. *Handb Clin Neurol* 2019; **163**: 147-164 [PMID: [31590727](https://pubmed.ncbi.nlm.nih.gov/31590727/) DOI: [10.1016/B978-0-12-804281-6.00008-2](https://doi.org/10.1016/B978-0-12-804281-6.00008-2)]
- 16 Arboix A, Bechich S, Oliveres M, García-Eroles L, Massons J, Targa C. Ischemic stroke of unusual cause: clinical features, etiology and outcome. *Eur J Neurol* 2001; **8**: 133-139 [PMID: [11430270](https://pubmed.ncbi.nlm.nih.gov/11430270/) DOI: [10.1046/j.1468-1331.2001.00180.x](https://doi.org/10.1046/j.1468-1331.2001.00180.x)]
- 17 Arboix A, Jiménez C, Massons J, Parra O, Besses C. Hematological disorders: a commonly unrecognized cause of acute stroke. *Expert Rev Hematol* 2016; **9**: 891-901 [PMID: [27367035](https://pubmed.ncbi.nlm.nih.gov/27367035/) DOI: [10.1080/17474086.2016.1208555](https://doi.org/10.1080/17474086.2016.1208555)]
- 18 Kloek AT, Seron MV, Schmand B, Tanck MWT, van der Ende A, Brouwer MC, van de Beek D. Individual responsiveness of macrophage migration inhibitory factor predicts long-term cognitive impairment after bacterial meningitis. *Acta Neuropathol Commun* 2021; **9**: 4 [PMID: [33407905](https://pubmed.ncbi.nlm.nih.gov/33407905/) DOI: [10.1186/s40478-020-01100-7](https://doi.org/10.1186/s40478-020-01100-7)]
- 19 Popp J, Bacher M, Kölsch H, Noelker C, Deuster O, Dodel R, Jessen F. Macrophage migration inhibitory factor in mild cognitive impairment and Alzheimer's disease. *J Psychiatr Res* 2009; **43**: 749-753 [PMID: [19038405](https://pubmed.ncbi.nlm.nih.gov/19038405/) DOI: [10.1016/j.jpsychires.2008.10.006](https://doi.org/10.1016/j.jpsychires.2008.10.006)]



Published by **Baishideng Publishing Group Inc**
7041 Koll Center Parkway, Suite 160, Pleasanton, CA 94566, USA

Telephone: +1-925-3991568

E-mail: bpgoffice@wjgnet.com

Help Desk: <https://www.f6publishing.com/helpdesk>

<https://www.wjgnet.com>

