Case Report

An 8-Year 5-Month-Old Boy with a Basal Ganglia Lesion with Triphasic Waves on Electroencephalogram

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Abstract

Background: Triphasic waves (TWs) on electroencephalograms (EEGs) have predominantly been observed in adults, often associated with Creutzfeldt–Jakob disease and metabolic encephalopathy. However, TWs have also been linked to various nonmetabolic and structural abnormalities. Additionally, reports of TWs in children are rare.

Case Presentation: We present the case of an 8-year and 5-month-old boy with basal ganglia lesion who exhibited TWs in the local C3 lead on electroencephalography. Subsequent EEGs revealed no additional abnormalities. During the follow-up at 1 year and 8 months, there was no significant change in the patient's condition.

Conclusion: Triphasic waves can occur in children with basal ganglia lesions, but their underlying causes may differ from those previously reported. Further research is needed to elucidate the mechanisms and clinical significance of TWs in pediatric patients.

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Keywords

triphasic wave; electroencephalogram (EEG); basal ganglia lesions; children; case report

Introduction

Electroencephalographic triphasic waves (TWs) are abnormal waveforms now recognized in the American Clinical Neurophysiology Society (ACNS) classification and revised glossary of electroencephalographic terminology and updated recommendations [1]. TWs consist of three distinct phases: a high-amplitude positive wave followed by a slow negative wave, occurring periodically at 1.5 Hz to 2 Hz [2]. They are typically bilaterally and widespread, with 60% predominantly in the frontal cortex and 40% posteriorly distributed or diffuse. Localized TWs have also been observed, especially in the frontal and central regions [3,4].

Foley *et al*. [5] initially reported that triphasic waves were observed in patients with hepatic encephalopathy and were described in 1950 as "blunted spikes and waves" in patients with hepatic encephalopathy. Subsequently, it was shown that this phenomenon can be detected in a variety [4] of metabolic encephalopathies, for example, Renal encephalopathy, hyponatremia, Creutzfeldt-Jakob disease, etc. [6]. Although the precise mechanism behind TW generation remains unclear, it is thought to be related to metabolic disorders. The specificity of TWs in etiological diagnosis remains debated. While some researchers believe that typical triphasic waves have diagnostic significance for

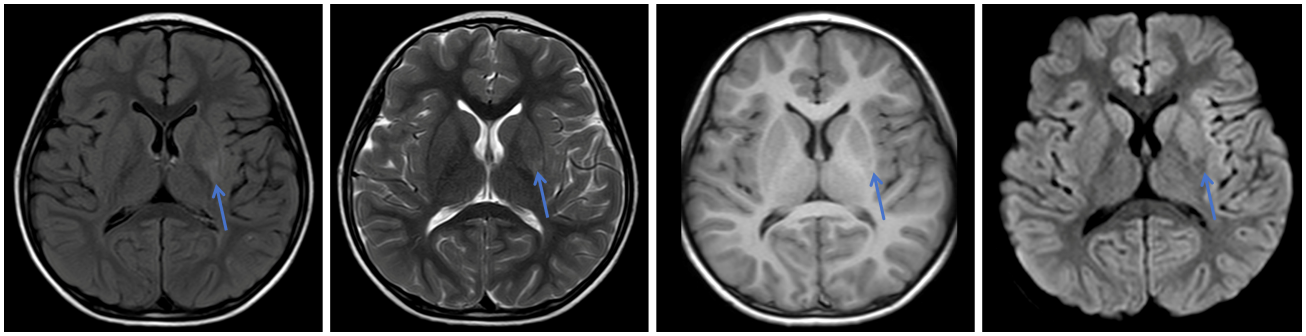

Fig. 1. Brain Magnetic Resonance Imaging (MRI) on February 4, 2022 (8 years and 5 months). Abnormal signal (blue arrow) in the posterior left lenticular nucleus: small abnormal signal on slightly high on T2 Weighted Imaging (T2WI), T2-Fluid Attenuated Inversion Recovery (T2-FLAIR), and T1 Weighted Imaging (T1WI), and slightly low on Diffusion Weighted Imaging (DWI), with unrestricted diffusion. (From left to right is T2W1, T2-FLAIR, T1WI, DWI.)

Hashimoto Encephalopathy (HE), most agree that typical and atypical TWs can appear in various toxic metabolic encephalopathies without distinguishing characteristics.

Triphasic waves are increasingly reported in diverse pathological conditions, predominantly in adults. However, these conditions are rarely observed in children, and the occurrence of TWs in pediatric electroencephalogram (EEG) recordings is seldom reported.

Case Presentation

Patient Information

An 8-year and 5-month-old male presented on February 4, 2022, with a two-month history of abnormal gait, accompanied by dizziness and hypoesthesia in the right foot. Over time, he developed blurred vision in his right eye and weakness in the middle, ring, and little finger of his right hand. There were no additional symptoms.

Clinical Findings

Physical examination revealed an abnormal gait, tiptoe walking, and limited dorsiflexion in the right fingers. The specialist physical test revealed the following: The muscle tone of the limbs was expected, the muscle strength of the right upper limb and right lower limb was level 4, the muscle strength of the left upper limb and left lower limb was level 5, the patellar reflex was slightly active.

Auxiliary Examination

An Magnetic Resonance Imaging (MRI) of the brain performed on February 4, 2022 (Fig. 1) revealed an abnormal patchy signal shadow in the left basal ganglia region. No abnormalities were detected on Magnetic Resonance Angiography (MRA) or Magnetic Resonance Venography (MRV). A follow-up MRI with contrast enhancement and spectral analysis on February 23, 2022 revealed abnormal signal shadows in the left basal ganglia, a slight increase in the Choline/Creatine (Cho/Cr) ratio, and a decrease in the N-acetyl-L-aspartic acid/Creatine (NAA/Cr) ratio, suggesting brain damage.

There were no abnormalities in blood ammonia, lactate, pyruvate/β-hydroxybutyric acid, thyroid function, complete blood transfusion immune system, erythrocyte sedimentation rate, blood α-fetoprotein (AFP), or human chorionic gonadotropin (HCG). Tests for central nervous system demyelination (conducted on February 19, 2022) and autoimmune encephalitis antibodies were negative.

Cerebrospinal fluid (CSF) analysis on February 18, 2022, showed no abnormalities in cell count, biochemistry, or smears. However, oligoclonal banding (Type II) was positive. Further CSF tests, including cytology, biochemical analysis, smear, fungal G, Galactomannan (GM) test, and tuberculosis infection T-cell assays, were all normal. CSF levels of HCG (2.5 mIU/mL) and AFP ($<$1.3 ng/mL), along with pathogen nucleic acids and tumor markers, tested on February 23, 2022, were all within normal ranges.

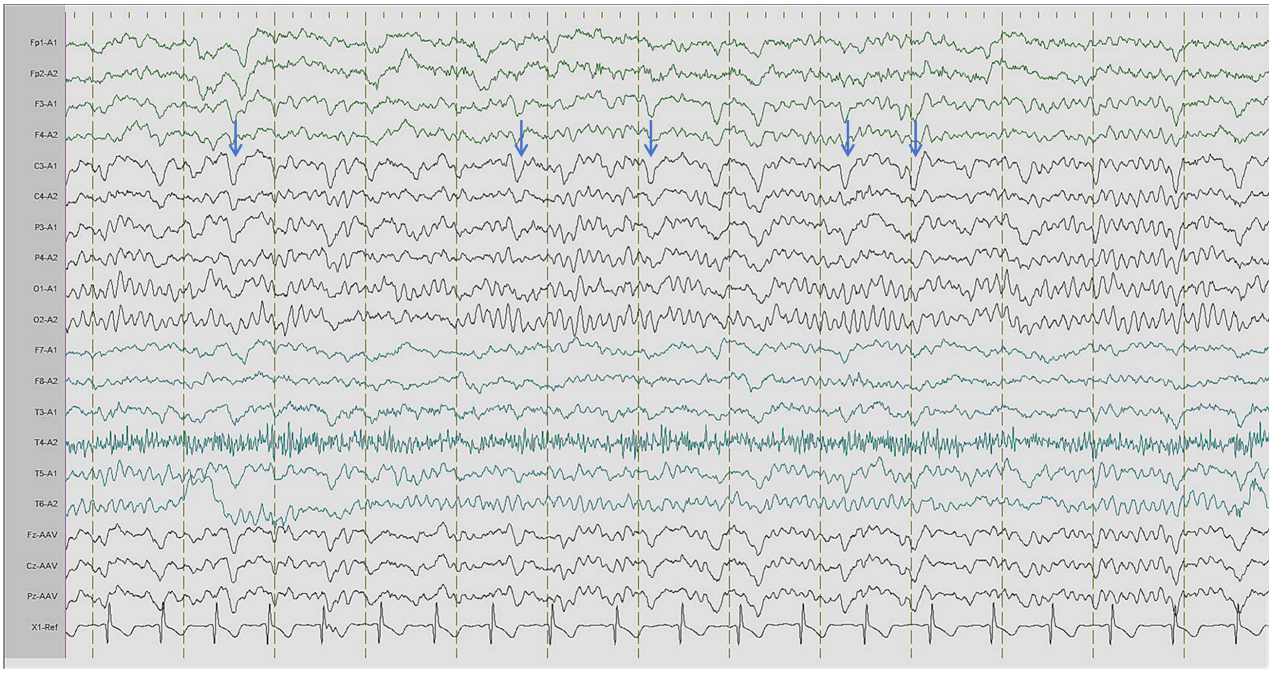

Fig. 2. EEG on February 16, 2022 (8 years and 5 months). Localized triphasic waves are visible in the C3, F3, and P3 leads, notably on C3. (EEG, electroencephalogram; blue arrow indicates triphasic waves.)

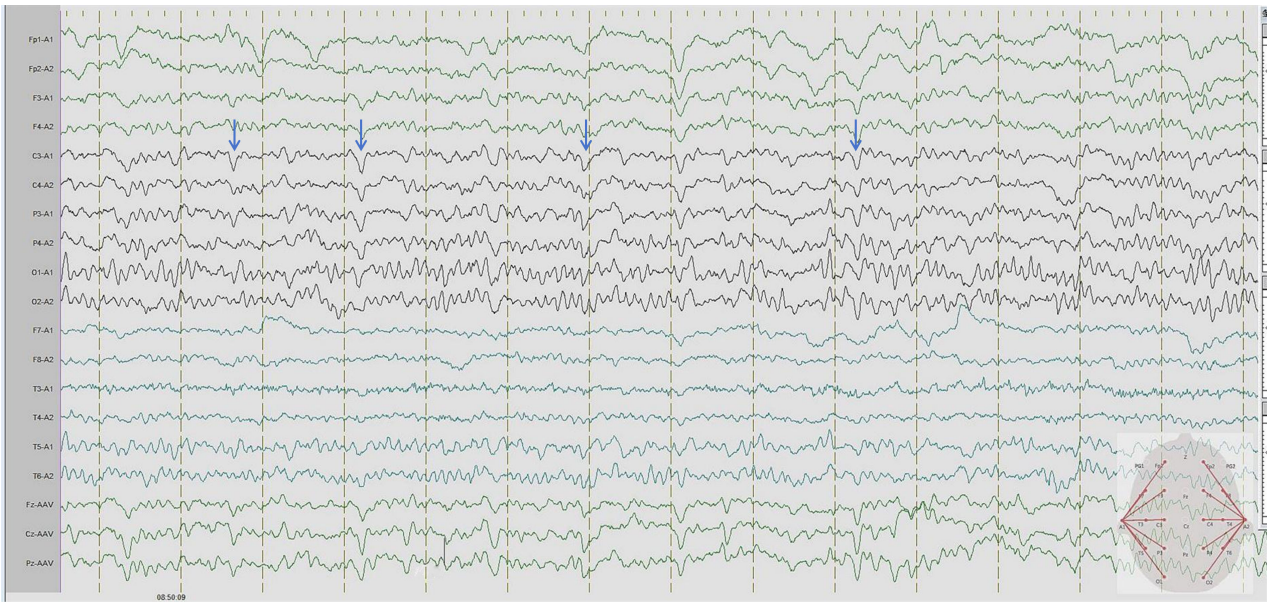

Fig. 3. EEG on July 22, 2022 (8 years and 10 months). Three-phase waves can be seen in lead C3. (EEG, electroencephalogram; blue arrow indicates triphasic waves.)

Diagnosis

The exact nature of the lesion in the left basal ganglia remains undetermined, though immune-related causes are highly suspected. The cerebrospinal fluid (CSF) showed oligoclonal banding (Type II), often associated with conditions like multiple sclerosis or autoimmune encephalitis. However, clinical and laboratory examinations do not support a diagnosis of multiple sclerosis. No abnormality in the head MRA and MRV makes the diagnosis of stroke unlikely. The four tests of demyelination of the central nervous system (Anti-Aquaporin 4 (Anti-AQP4) antibody,

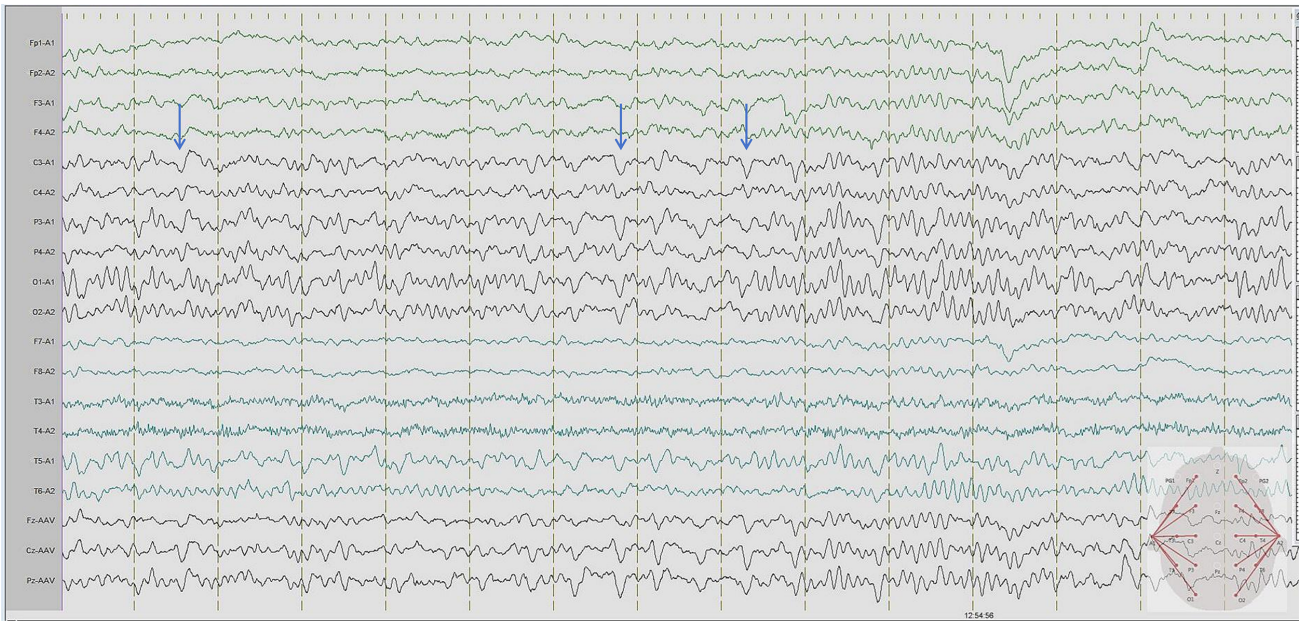

Fig. 4. EEG on April 25, 2023 (9 years and 7 months). Three-phase waves were visible in the lead C3. (EEG, electroencephalogram; blue arrow indicates triphasic waves.)

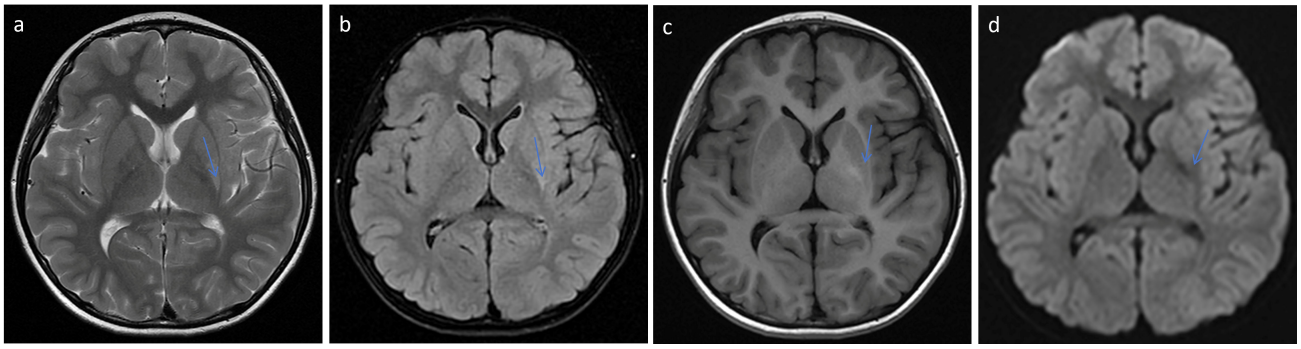

Fig. 5. Brain MRI on July 22, 2022 (8 years and 10 months). a, T2WI; b, T2-FLAIR; c, T1WI; d, DWI. Small abnormal signal (blue arrow) in the left posterior lenticular nucleus on (d, DWI), slightly high on T2WI, T2-FLAIR, and T1WI, slightly low on DWI, with unrestricted diffusion. Compared to February 4, 2022, the area of abnormal signal in the left posterior portion of the lenticular nucleus reduced.

Anti-myelin oligodendrocyte glycoprotein (Anti-MOG) antibody, Anti-Myelin basic protein (Anti-MBP) antibody, Anti-Glial Fibrillary Acidic Protein (Anti-GFAP) antibody) are negative. Inflammatory demyelinating disease of the central nervous system is not supported.

Inflammation and infection are considered improbable due to the lack of abnormalities in related examinations. There is no abnormality in the examination of cerebrospinal fluid HCG, AFP and tumor markers, and the lesions in the left basal region have a tendency to reduce in a follow-up brain MRI, which does not support the diagnosis of tumors. The patient received treatment with B vitamins, Micoba, and Xinkelai for neuroprotection, though the efficacy of these interventions remains uncertain. At 1 year and 8 months follow-up, there was no significant change in the condition of the patient.

Timeline

The patient underwent three routine electroencephalogram examinations at 8 years and 5 months, 8 years and 10 months, and 9 years and 7 months. The international 10–20% system standard 19-lead was used to collect the awake electroencephalogram for 30 minutes. The first C3 lead was visible. Localized triphasic waves can be seen in

C3-A1, F3-A1, and P3-A1 leads, as shown in Fig. 2. The second and third EEG signals showed triphasic waves in lead C3, as shown in Figs. 3,4. Brain MRI was reviewed on July 22, 2022 (8 years and 10 months, Fig. 5). On T2 Weighted Imaging (T2WI), T2-Fluid Attenuated Inversion Recovery (T2-flair), T1 Weighted Imaging (T1WI), and Diffusion Weighted Imaging (DWI), there are abnormally small signals at the left posterior part of the lens nucleus (blue arrow), slightly higher signals at T2 Weighted Imaging (T2WI), T2-flair, T1WI, and slightly lower signals at DWI, with unrestricted diffusion. Compared to February 4, 2022, the area of abnormal signals at the left posterior part of the lens nucleus is smaller than the former.

Discussion

Triphasic waves (TWs) on electroencephalograms (EEGs) have primarily been observed in adults, with limited reports in children. We present the case of an 8-year and 5-month-old boy with basal ganglia lesion who exhibited TWs localized to the C3 lead on EEG. No other abnormalities were detected across A series of EEGs.

Basal ganglia lesions are typically associated with degenerative diseases but can also arise from cerebrovascular disease, inflammation, poisoning, or tumors. In the present case, the lesion is likely immune-related. Given the deep location of the basal ganglia, imaging is crucial for diagnosis. Previous experience has suggested that electroencephalography (EEG) does not play an important role in the diagnosis and treatment of simple basal ganglia lesions.

A three-phase electroencephalogram wave is a relatively special periodic abnormal waveform. It is often closely related to certain diseases and has suggestive significance in clinical diagnosis and treatment. TWs are often associated with metabolic encephalopathies, especially hepatic encephalopathies. In addition, Angelman syndrome may exhibit TWs in their EEG during seizures. However, recent case reports have shown that TWs can be secondary to other conditions beyond metabolic encephalopathies. These include electrolyte imbalances, such as uremia, hypernatremia, hyponatremia, hypercalcemia, and hyperammonemia [7]; endocrine disorders like hypoglycemia, hyperthyroidism, hypothyroidism, and isolated ACTH deficiency; as well as drug toxicity, particularly from lithium [8]; and hypoxic encephalopathy. TWs have also been linked to stroke, tumors, Creutzfeldt-Jakob disease, autoimmune encephalitis, dementia [9,10], and low intracranial pressure [11]. Inflammatory and infectious encephalopathies, such as sepsis, Mohs meningitis, herpes simplex encephalitis, Borrelia burgdorferi meningoencephalitis, tuberculous meningitis, carcinomatous meningitis, and Hashimoto's encephalopathy, have also been associated with TWs [12–14].

While TWs are common in adults, they are relatively rare in children. Janati and Erba [15] reported TWs in two pediatric drowning cases, which involved repetitive focal biphasic or triphasic waveforms. Ke Zhang *et al.* [16] described a case of anti-N-methyl-D-aspartate (anti-NMDA) encephalitis with TWs in a 9-year-old girl, and Hosain *et al.* [17] found TWs in six out of 178 comatose children, suggesting metabolic encephalopathy. Laan *et al.* [18] identified TWs in children with Angelman syndrome, a rare genetic disorder. Given the broad range of causes, metabolic encephalopathy is often considered first, but in our case, no metabolic abnormalities were identified, effectively ruling out this possibility. TWs have also been reported in toxic encephalopathies caused by medications such as valproic acid, cefepime, cefoperazone, ceftriaxone, lithium, and pregabalin [19,20]. Vulliemoz *et al.* [21] also reported a case of TWs in a patient with chronic renal failure after levetiracetam. However, none of these factors were present in our patient, making drug-induced EEG abnormalities unlikely.

The mechanism underlying TWs remains unclear. Bickford and Butt [22] proposed that TWs may result from conduction waves along the cortex triggered by subcortical disorders at the thalamocortical level. Gloor P and Fariello RG [23] suggested that abnormal oscillatory firing between cortical and thalamic neurons could be responsible for this EEG pattern. Currently, TWs are thought to arise from metabolic or structural abnormalities in thalamocortical relay neurons, with abnormal glutamate metabolism potentially playing a role. In our case, the electroencephalogram of the child showed TWs associated with basal ganglia lesion, but the precise mechanism remains unknown and warrants further investigation.

The prognosis of patients with TWs varies. Bahamon-Dussan *et al.* [24] studied 30 patients with TWs, reporting a 77% mortality rate after 22 months of follow-up, with only three of the seven survivors being neurologically normal. They concluded that the prognosis of patients with TWs is poor [24]. Sutter *et al.* [25] analyzed EEGs from 154 patients with encephalopathy, finding that TWs were associated with increased mortality and adverse outcomes. Indeed, reports have also mentioned that patients exhibiting TWs have experienced favorable prognoses following active treatment [12,13]. Research reveals that TWs can be observed in a wide range of diseases, making it nonspecific in diagnosing the underlying cause [26]. Consequently, the

prognosis of patients with TWs is likely to depend on the treatability of the fundamental disease that generates them, rather than the presence of the TWs themselves.

In our patient, the primary condition was a basal ganglia lesion with uncertain etiology, with no identifiable cause associated with TWs. After more than one year of follow-up, the condition of the patient remained stable. Continued monitoring will be necessary to assess the progression of the disease.

Conclusion

Electroencephalograms (EEGs) in children with basal ganglia lesions may reveal localized or sporadic TWs. The prognosis of this patient remains uncertain, necessitating continued follow-up. While TWs are commonly associated with conditions such as metabolic encephalopathy and Creutzfeldt-Jakob disease, they are not specific markers. Clinicians should consider a broader differential diagnosis when encountering TWs on EEG. To our knowledge, this is the first report of TWs in a child with basal ganglia lesions.

Availability of Data and Materials

The data supporting the findings of this study are included in the article. Further inquiries can be directed to the corresponding author.

Author Contributions

YLZ was responsible for the research study design, project execution, whole-case analysis and manuscript preparation. TY provided EEG analysis. JNC provided project execution, analyzed the EEG and MRI data, and Visualize the results. QYF provide the research study design, project execution and the interpretation of the data. GN provided MRI images and the related images analysis. RL provided the research study design, clinical guidance, the project supervision, and offered solutions to challenges encountered during the research. YLZ drafted this manuscript. All authors contributed to important editorial changes in the manuscript. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics Approval and Consent to Participate

This study was conducted in accordance with the principles of the Declaration of Helsinki and was approved by the Ethics Committee of West China Second Hospital of Sichuan University (approval number: 2023-271). The ethics committee granted exemption from informed consent.

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Conflict of Interest

The authors declare no conflict of interest.

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