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Clinical Evolution of a Case of Sydenham's Chorea under Sodium Valproate: A Case Report Followed at the University Clinics of Bukavu

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Abstract

Sydenham's chorea is a post-streptococcal complication that is still common in developing countries, often linked to poorly or untreated recurrent tonsillitis. We report the case of a 17-year-old female patient presenting with sudden choreic movements after a febrile episode, with a history of recurrent tonsillitis. Initial treatment with phenobarbital proved ineffective, leading to the introduction of sodium valproate at a dose of 750 mg twice daily. The outcome was favorable with complete disappearance of abnormal movements within the first few days and no recurrence at one month of follow-up.

Keywords

Sydenham's Chorea, Recurrent Tonsillitis, Sodium Valproate, Case Report

1. Introduction

Sydenham's chorea is an autoimmune neuropsychiatric disease and is considered a major manifestation of acute rheumatic fever. It is becoming the leading cause of acquired chorea in children in the Third World [1].

It results in involuntary movements, linked to the contraction of several muscles. These movements are sudden, brief, rapid, unpredictable, and of variable amplitude, often generalized and can affect a region of the body, in particular the limbs, the face, etc. and are associated with psycho-characteristic disorders [2].

Regarding the age of onset, the literature generally reports an average age ranging from 5 to 15 years. In our case, we observed a slightly higher age of 17 years, which exceeds the average age reported by MBAYE K. *et al.* (2022) in their study conducted at the ALBERT ROYER National Children's Hospital, where the mean age of patients with Sydenham's chorea was 9.45 years, with extremes ranging from 8 to 15 years [3]. It is of variable etiology [4].

It is caused by group A beta-hemolytic streptococcus. The mechanism is probably autoimmune, secondary to a cross-reaction between antibodies directed against the M protein of streptococcus A and neurons of the central gray nuclei [1].

Classic management of Sydenham's chorea combines antibiotic prophylaxis of acute rheumatic fever (ARF) and Haloperidol [4].

Abnormal movements often pose diagnostic and therapeutic difficulties [5].

RAA and Sydenham's chorea remain a major public health problem in developing countries due to diagnostic difficulties and high cost of management [6].

This is how we considered it useful to present the clinical, therapeutic and evolutionary profile of post-streptococcal chorea in a patient followed in the internal medicine department of the University Clinics of Bukavu.

2. Case Presentation and Observation

This is a 17-year-old patient, living in BURIBA in the commune of BAGIRA, South Kivu Province in the Democratic Republic of Congo, with an average socioeconomic level, who came to consult us for holocranial, frontal headaches rated at 7/10. on a digital scale and fevers without a reported schedule and whose onset dates back to 24 hours of our consultation. She allegedly self-medicated with Paracetamol tablets of 500 mg twice a day but without success. The evolution was marked by the sudden onset of abnormal bilateral choreic movements and this is what also motivated the present consultation at the University Clinics of Bukavu for better management.

There is a history of recurrent angina treated intermittently.

Physical examination on admission found a general condition marked by an active bedridden attitude and a stage 1 coma (obnubilation). The axillary temperature was 35.6˚C, blood pressure 141/89 mmHg, with regular tachycardia at 127 beats per minute, polypnea at 26 cycles per minute, SaO_2 at 99% in free air.

On oropharyngeal examination we noted redness of the oropharyngeal mucosa and a neurological examination marked by obtundation. Abnormal, sudden choreic movements of the face and upper limbs on motor examination.

The paraclinical examinations were normal with the ASLO normal at 74 IU/ml. The ASDOR, DNAses, not done, a blood count with a hemoglobin at 14.9 g/dl and normal white blood cells at 7200 cells/field. The CRP normal at 4.34 mg/L. The throat swab culture is not done. The cardiac ultrasound and the ECG without particularities noted.

The diagnosis of Sydenham's chorea outside of acute articular rheumatism, in

a context of repeated angina was made.

In view of this condition, the patient was hospitalized and put on injectable Phenobarbital 100 mg in 5% glucose serum, 500 CC to be made to flow for 4 hours then Phenobarbital tablet 100 mg 1 time 1 tablet, injectable Augmentin 3 times 1.2 grams per day for ten days. The response to Phenobarbital was marked by the regression of abnormal movements a few minutes after administration of the drug.

We also note the reappearance of abnormal chorea-type movements on the second day of treatment, occurring in a paroxysmal manner.

This motivated us to stop the phenobarbital and put the patient on oral sodium valproate at 750 mg twice daily.

The evolution was marked by the disappearance of abnormal movements during the entire duration of her hospital stay and an absence of fever. After 7 days of treatment with sodium valproate, our patient was discharged. A check-up was carried out after 1 month of discharge and we did not note a reappearance of abnormal movements (Table 1).

After 3 months she returned for follow-up, her general condition was preserved, all vital signs within normal limits and without any reported abnormal movements. Since her discharge until today, it is already more than 6 months without abnormal movements prompting her to come back for a consultation.

Table 1. Summaries of the clinical and therapeutic chronological evolution of the case

Events	Deadline or approximate date
Last episode of tonsillitis	Not dated precisely, but several months earlier according to the interrogation
Appearance of chorea	24 hours after the onset of fever and headache
Admission to hospital	The day the abnormal movements appeared
Initiation of Sodium Valproate	The second (2nd) day of hospitalization, after failure of Phenobarbital
Discharge from hospital	Seven (7) days after the start of treatment with Sodium Valproate
Follow-up visit	- First visit: 1 month after discharge - Second visit: 3 months after the first visit

3. Discussion

Sydenham's chorea, formerly called St. Vitus' dance, is the leading cause of acquired chorea in developing countries [4]. It follows a repeated group A Streptococcal infection during early life. A correlation of a low socio-economic level is associated with it. Well-conducted antibiotic therapy for the treatment of tonsillitis, and the rise in socio-economic and health levels have significantly reduced the prevalence of Sydenham's chorea in developed countries [6].

For this study, it is a 17-year-old patient.

Starting from the female gender, this result corroborates with several data found in the literature, notably in a study conducted in Morocco on "Post-streptococcal chorea at the CHU Marrakech Children's Hospital" shows that out of the

7 cases followed, 4 cases were female, or 57.1% [2]. This female predominance was noted in another study conducted in Senegal on 11 cases followed, 8 of which were female, or 72.72% [3]. However, a few rare series report a male prevalence [1].

Based on the age of onset, the literature suggests an average age that varies between 5 and 15 years; A difference of two years is observed in our patient, *i.e.* 17 years, an age higher than that found by MBAYE K. at al who notes that in their study the average age was 9.45 years and extremes ranging from 8 to 15 years [3]. Furthermore, the age of our patient is close to that found by S. BOUCHAL *et al.* on "Spectacular response to sodium valproate of recurrent Sydenham chorea" from 2014, in a 16-year-old girl [4].

The history of repeated and insufficiently treated angina is noted in this patient without diagnosis of ARF. These elements find support in several cases found in the literature.

In our study, the onset was marked by a feverish state and after 24 hours, abnormal movements localized to the upper limbs suddenly appeared, abrupt and choreic, after several episodes of angina noted during questioning. This result, according to the mode of occurrence, corroborates that found by I. JELLAB *et al.*, 2009, suggesting a progressive and insidious onset following streptococcal angina 1 to 6 months [2].

Chorea is usually generalized but localized and bilateral in this patient; this predominance was reported however by AI-Eissa [1].

Initially our patient was put on Phenobarbital in combination with Amoxicillin-Clavulanic acid with a poor response since a reappearance of abnormal movements 24 hours later. Furthermore, in the literature haloperidol is the most widely used neuroleptic in the treatment of Sydenham's chorea with good results as has been noted in different series in the literature [4]. Its tolerance is however not always good, drowsiness under haloperidol can be observed.

Based on the response to sodium valproate, we observed a spectacular response from the start of this treatment with a definitive cessation during her entire hospital stay. This result corroborates with that found in another study conducted by S. BOUCHAL *et al.* showing a spectacular result in a 16-year-old girl followed under sodium valproate [4].

In the context of this clinical picture involving choreic movements in an adolescent girl, two important differential diagnoses were considered: Huntington's disease and autoimmune encephalitis, particularly anti-NMDA receptor antibody encephalitis.

Huntington's disease, an autosomal dominant hereditary neurodegenerative disorder, typically presents with progressive involuntary movements, cognitive impairment, and psychiatric disorders. It rarely begins before adulthood, except in its juvenile form [7]. In our study, the absence of a similar family history, the isolated age of onset at 17 years without associated cognitive or psychiatric signs, and the complete reversibility of symptoms under treatment, excluded this diagnosis. In addition, the rapid and favorable evolution under sodium valproate is

atypical for a neurodegenerative disease (Table 2).

Concerning autoimmune encephalitis, in particular anti-NMDA encephalitis, it can present in adolescents with abnormal movements, but is generally associated with a marked psychiatric picture, epileptic seizures, dysautonomia, or a marked alteration of the state of consciousness [8] [9]. In our observation, the absence of profound confusional syndrome, vegetative instability, as well as the context of untreated recurrent angina in a region with a high endemicity of ARF, rather point towards Sydenham's chorea. Finally, the spectacular evolution under sodium valproate and the absence of relapse during follow-up also support a benign post-infectious origin rather than a severe autoimmune one (Table 2).

Table 2. Different features between Sydenham's chorea, Huntington's disease and autoimmune encephalitis (anti-NMDA).

Features	Sydenham's chorea	Huntington's disease	Autoimmune encephalitis (anti-NMDA)
Age of onset	5 to 15 years (exceptional after 18 years)	30 - 50 years (rare juvenile form, before 20 years)	Children, adolescents and young adults
Onset of symptoms	Sudden or subacute, often after an infectious episode	Progressive over several months	Subacute, within days to weeks
Infectious or inflammatory context	Yes, post-strep throat	No	Sometimes preceded by a viral infection
Family history	Often absent	Yes, (autosomal dominant, HTT mutation)	Nonspecific, but sometimes associated with a tumor
Motor symptoms	Generalized, abrupt, non-rhythmic choreic movements	Progressive choreic movements, dystonias, rigidity	Facial dyskinesias, chorea, orofacial movements
Psychiatric symptoms	Sometimes: irritability, emotional lability	Yes: cognitive disorders, dementia	Marked: agitation, hallucinations, behavioral disturbances
Cognitive disorders	Absent or discreet, transient	Progressive and severe in the long term	Common, associated with confusion or mutism
Associated neurological signs	Mild coma possible at the beginning, without focal deficit	Cognitive decline, extrapyramidal signs	Seizures, impaired alertness and catatonia
Biological assessment	High ASLO, sometimes high ESR and CRP	Non-specific	Pleocytosis, specific antibody in CSF
Brain MRI	Normal or non-specific	Gray nuclei atrophy (advanced stage)	Possible frontal or temporal anomalies
EEG	Normal or non-specific	Not very useful	Diffuse slowdown, extreme delta brush activity
Evolution without treatment	Spontaneous favorable or recurrences	Progressive, incurable deterioration	Severe without treatment, potentially fatal
Response to Sodium Valproate	Excellent (often spectacular)	Little or no effect	Not very effective alone, immunomodulatory treatment necessary
Specific treatment	Antibiotic prophylaxis, sodium valproate, neuroleptics	No curative treatment, symptomatic management	Immunotherapy (corticosteroids, IgIV).

4. Conclusions

Sydenham's chorea is a first acquired chorea, although very rare in developed countries and the advent of antibiotic therapy in the management of RAA has largely contributed to its disappearance.

However, the diagnosis of Sydenham's Chorea is always made clinically, in a context of repeated angina; poorly treated or insufficiently treated in developing countries.

Several treatments are current; in our patient, sodium valproate gave good results, marked by the complete regression of abnormal movements from the introduction of treatment.

Authors' Contributions

All authors contributed to this work and have read it.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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