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SHORT COMMUNICATION

Somatic symptom disorder should be suspected in children with alleged chronic Lyme disease

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Abstract

We report a case series of seven children admitted to a tertiary level pediatric ward for long-lasting physical symptoms with a previous diagnosis of chronic Lyme disease. In these children, medical history and clinical features were strongly suggestive of a psychopathological disorder, mainly a somatic symptom disorder.

What is Known:
- *There is an increasing number of diagnoses of chronic Lyme disease both in North America and in Europe. Adults receive this diagnosis to explain chronic physical complaints often with negative history and serology.*

What is New:
- *Somatic symptom disorder should be suspected in children and adolescents with non-specific symptoms diagnosed with chronic Lyme disease.*

Keywords Chronic Lyme disease · Somatic symptom disorder · *Borrelia burgdorferi* · Doctor shopping

Abbreviations
CLD Chronic Lyme disease
SSD Somatic symptom disorder

Background

Chronic Lyme disease (CLD) is a diagnosis used both in North America and in Europe to label patients with chronic physical complaints, such as pain, fatigue, fever, and neurocognitive symptoms, irrespective of clinical or serologic evidence of previous Lyme disease [1, 2].

CLD symptoms do not fit into any of the recognized diagnostic categories of Lyme disease such as cutaneous involvement, arthritis, aseptic meningitis, facial nerve palsy, encephalitis, polyneuritis, uveitis, and carditis. There is very little evidence to suggest that CLD is recognized by any scientific society and no established diagnostic criteria are available [3]. Nevertheless, people may often be diagnosed or self-diagnose it [4].

Studies in adults highlighted how, behind this alleged CLD, may hide a psychopathological disorder, but no data are available in children [5].

Somatic symptom disorder (SSD) is defined, according to the fifth edition of the Diagnostic and Statistical Manual of

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Mental Disorder (DSM-V) [6], by the presence of nonspecific discomforting somatic symptoms (mostly head, abdominal, back, and muscle pains) and excessive thoughts on a possible disease, that seriously undermine children daily life causing in severe cases school absenteeism, loss of interests and activities, isolation from pairs, abandonment of sports activities, for more than 6 months. The attention focused on somatic symptoms, the repeated request for help, and the avoidance of physical activity strongly support the diagnosis of SSD.

SSD is one of the most frequent psychopathological disorders in children and adolescence, with a prevalence reported as high as 15% of adolescents in outpatient setting and a prevalence of 8.6% of causes of non-traumatic pain in a series of children and adolescents referring to a pediatric emergency department [7], with an increasing trend in high-income countries [8]. SSD can be triggered by different stressors (e.g., catastrophic family events, abuse, bullying, gender dysphoria), may have a severe impact on the quality of life, and is associated in 30% of cases with psychiatric co-morbidity such as anxiety disorder and depression [9].

Evidence shows that patients with SSD often receive another diagnosis before a correct SSD one [7].

The aim of this study was to investigate the numbers of pre-admission CLD diagnosis in a population of children admitted to a tertiary level pediatric ward, as well as their clinical features.

Methods

This was a retrospective study conducted in the pediatric ward of the tertiary level, university teaching, children's hospital, Institute for Maternal and Child Health, IRCCS Burlo Garofolo of Trieste, Italy. The study protocol was approved by the institutional review board.

The electronic medical records of all patients admitted between January 2016 and December 2018 were reviewed. Initially, we selected all medical records in which the term "Lyme" was present, then we focused on the records in which the medical history was remarkable for a diagnosis of CLD. For every selected patient, we collected information about age, gender, geographic origin, medical history, symptoms, diagnostic tests, specialist consults, previous hospitalizations, and final diagnosis at discharge. All the information about socio-psychological aspects and markers of children's "loss of function" such as days of absence from school related to the symptoms, isolation from pairs, avoidance of physical activity, loss of interests, and hobbies were proactively searched and registered as part of a standard approach to a positive SSD diagnosis. This is part of an Institutional protocol in all suspect cases starting from the emergency department evaluation [7] by the attending pediatrician.

Each patient was evaluated by a child neuropsychiatrist during admission and one of them also at follow-up. In the Italian Public Health System, there is no formal distinction between child psychiatrist and child neurologist and a doctor with a post-graduate degree of *Neuropsichiatria Infantile* takes care of patients with both neurological and psychiatric disorders till the age of 18. Child psychologists are involved in diagnosis and care by the neuropsychiatric consultant.

Results

From January 2016 to December 2018, 2552 patients were admitted. Through the analysis of the electronic medical records, we found that the term "Lyme" was present in the records of 26 patients. After reviewing these records, we found seven patients who had received before admission the diagnosis of CLD.

Table 1 shows the main features of these patients. In summary, they were adolescents, living in an endemic area for Lyme disease, with long-lasting physical symptoms. The most recurrent symptoms were headaches that affected five patients out of seven, specifically a tension headache, reported low-grade fever in four patients, which was not recorded during hospital stay, and arthralgia without signs of active inflammation, in three patients. Five patients also complained of vague asthenia, not linked to physical efforts. One patient reported difficulty in walking so as to arrive at the use of a wheelchair and one visual disturbance.

Specific psychosocial risk factors were identified in four patients, respectively: high school and family demanding with perfectionism features, the sudden death of a parent, a history of bullying, and gender dysphoria.

All the patients had received the diagnosis of CLD for the same symptoms that brought them to admission. The diagnosis of CLD was made mainly by physicians, but in one case it was self-made by parents. Before the diagnosis of CLD, the serology for *Borrelia burgdorferi* was sought in all cases (both ELISA and Western blot): IgM were positive in 2 patients, while IgG were negative in all patients. After the diagnosis of CLD, all patients received cycles of antibiotic therapies without resolution of symptoms. Due to their symptoms, they underwent a considerable amount of diagnostic tests, specialists' evaluations, and hospitalizations. Symptoms strongly impaired their school attendance. Reviewing medical history and clinical features, the diagnosis of a previous Lyme disease was not confirmed in any patient. On the contrary, a diagnosis of a psychopathological disorder, mainly SSD, was made in six patients after a joint evaluation by a pediatrician and a child neuropsychiatrist according to the DSM-V criteria.

The non-specific symptomatology behind which disproportionate thoughts and excessive health concerns were hidden was found to have a negative impact on the quality of life, leading also to school dropout. In five patients out of seven, symptoms lasted more than 6 months.

Table 1 Clinical features of children with a previous diagnosis of CLD at admission and final diagnosis

	Age	Gender	Symptoms	Duration of symptoms	Days of school lost in the 3 months before	Investigations already performed**	Number of hospitalizations	Who suspected diagnosis of CLD	Serologic tests for Lyme before diagnosis of CLD	Demonstration of previous Lyme during admission	Final diagnosis
1	14 years	F	Mild fever*, headache, asthenia	3 months	15 days	Blood test, blood culture, urine culture, Mantoux, Quantiferon, chest X-ray, echocardiography, brain MRI	2	GP	IgM positive/IgG negative	No	Viral infection
2	13 years	F	Arthralgia, mild dysarthria, not walking	More than a year	Reduced school hours, only 2.3 days/week for 1 year	Blood test, otolaryngology visit, brain MRI, SSEP, MEP, knee MRI	3	Hospital	IgM/IgG negative	No	Anxiety disorder with somatic complaints
3	14 years	M	Arthralgia, asthenia, headache, memory deficit, fever*	1 year	1 month	Blood test, rheumatological and psychiatric consult	2	GP	IgM positive/IgG negative	No	Somatic symptom disorder
4	16 years	F	Mild fever*, occasional headache, asthenia	Almost a year	1 month	Blood test, abdomen ultrasound, chest X-ray and CT, eye examination	2	Patient's parents	IgM/IgG negative	No	Somatic symptom disorder
5	13 years	M	Fever*, headache	More than a year	2 months	Blood test, abdomen ultrasound, chest X-ray, rheumatological and pulmonary consult	2	GP	IgM/IgG negative	No	Somatic symptom disorder
6	12 years	F	Abdominal pain, arthralgia, headache with transient photophobia	More than a year	2 months	Blood test, esophagogastroduodenoscopy, abdominal X-ray, EEG, brain MRI	5	Infectivologist	IgM/IgG negative	No	Somatic symptom disorder
7	17 years	M	Asthenia, atopic dermatitis	6 months	School dropout	Blood test, infectivological and pulmonary consult	3	Consultant in private practice	IgM/IgG negative	No	Somatic symptom disorder

Blood test—blood count cell, liver and kidney functioning, serology (IgM/IgG *Borrelia burgdorferi*)

*Fever was always reported and never documented during hospitalizations

**Before our admission

Discussion

This study shows that in pediatric patients with chronic physical symptoms a diagnosis of CLD may hide a psychopathological disorder, mainly a SSD.

We report on seven adolescents with long-lasting physical complaints, which were already labeled as affected by CLD. This diagnosis was mainly based on non-specific symptoms with an isolated IgM positivity for *Borrelia burgdorferi* in only two cases. In this sense, as reported in the literature, it should be emphasized that the positive predictive value of serologic testing in children living in endemic areas for Lyme disease is low, with a well-known risk of false-positive test results [10, 11]. We suggest that serologic testing should not be routinely performed in children with non-specific subjective physical complaints and that the tests' results should be strictly related to clinical features specific of Lyme disease.

The concept of CLD is a matter of discussion in the scientific community, and with this study, we did not aim to investigate its existence as a real clinical entity. Nevertheless, this study found that, at least in an endemic area for Lyme disease, children with long-lasting unclear symptoms may end up running diagnostic tests for Lyme disease or directly receive a diagnosis of CLD by physicians or even by parents, based on information derived from the web and social media. Remarkably, in this series, children with an alleged CLD did not resolve their symptoms despite cycles of focused therapies and families were endlessly searching a medical explanation for their complaints, thus leading to hospitalization in our ward.

Medical history and clinical features were similar in characteristics, highly suggestive for SSD: long lasting physical complaints, a detrimental impact of symptoms on daily activities, an extensive diagnostic workup and "doctor shopping" performed before diagnosis [11, 12] without any objective or laboratory sign of disease.

From an epidemiological perspective, SSD is more frequent in adolescence than Lyme disease, even in endemic areas. It is well known that the time between the onset of symptoms and diagnosis of SSD impacts on prognosis [7, 12]. Therefore, in children affected by long-lasting physical complaints with a strong impact on daily activities, without a clearly identified cause, SSD should be promptly suspected. In this sense, a diagnosis of CLD may be detrimental, both leading to unjustified and potentially harmful treatments and delaying a possible diagnosis of SSD, limiting the chance of these patients to receive a timely proper support.

This is a retrospective study with some limitations. We collected a relatively small sample from a single institution, and we may have missed some patients who received a diagnosis of CLD. Follow-up after discharge to assess patients' outcomes was not available for all patients.

Lastly, being performed in an endemic area for Lyme disease, we cannot generalize our findings to other geographical areas.

In conclusion, this study shows that, as suggested for adults [5], children and adolescents labeled as affected by CLD may have a psychopathological disorder.

Author contributions F.P., D.N. and G.C. had primary responsibility for writing the manuscript.

F.P., D.N., A.V., G.M. and A.O. were responsible for data collection.

E.B. has supervised and contributed to the writing of the manuscript.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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