Illness trajectories in Mexican children with juvenile idiopathic arthritis and their parents

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Introduction

Juvenile idiopathic arthritis (JIA) is a group of chronic diseases of children and adolescents characterized by pain and joint swelling, limited mobility and functional disability in the inflammatory phase as well as structural damage and functional impairment throughout the years [1, 2]. Likewise, various extra-articular manifestations, comorbid conditions and drug adverse events may occur. In consequence, JIA may influence children’s growth and their physical, psychological, educational, economical and social development [3]. In this context, the quality of life (QoL) of JIA children and close relatives may be deeply affected by the disease [3–5].

Expectations for children with JIA and their families vary across nations. In developed countries, children are more likely to be diagnosed and treated early on in the course of JIA than in developing countries, where they are often misdiagnosed and treated by laypeople or even physicians for whom JIA is unknown; by the time these children receive proper care, the disease has already affected their functioning, QoL, expectations, therapeutic compliance and efficiency of available treatments [6, 7].

In general, the approach of JIA children’s QoL has focused on the quantitative assessment of health-related quality of life (HRQoL) by using generic and disease-specific instruments that, despite their value, have some limitations. Mainly HRQoL does not include most of the cultural aspects of the children’s life and the meaning of QoL, and validity of instruments differ across different populations.

In this study, we were interested in the meanings that parents and children have of their experiences with JIA and the surrounding world. Therefore, we considered that the qualitative approach—which essentially aims to explain the socio-cultural world through the eyes of individuals [8, 9]—would give us a wider and better scope of the impact of JIA. In this context, we hypothesize that by knowing the role of socio-cultural aspects, particularly the meaning and impact of JIA, we could enrich the patient–physician relationship, therapeutic efficiency and quality of care.

Theoretical framework

This study was conceived through the microlevel perspective of Medicinal Anthropology—or Medical Anthropology—and specifically of Clinical Anthropology [10–13]. Culture was defined as the knowledge learnt and shared by a group of individuals living in a society that based its behaviors, changes or adaptations according to circumstances [11]. Part of this definition is the people’s way of living: their beliefs, values, communication, habits

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and customs. Society, on the other hand, refers to the group of individuals interacting in a geographical area in accordance to its own culture [11, 14].

Theoretical position

The theoretical position of this study was based on three complementary theories: suffering [13], explanatory models [15] and illness trajectory [16, 17]. We gathered and analysed patients’ and parents’ narratives in order to reconstruct illness trajectories, recognize the cultural context in which the patient was set, and interpret his experience and meaning of disease. Disease was defined as the diagnosis made by a professional of medicine [15, 18]. Illness corresponded to the form in which the patient defined his unwell health status and the cultural dimensions of disease, particularly when referring to the semiotic, semiological, and phenomenological construction of symptoms [13, 15, 18].

The theory of suffering refers to the meaning of disease and aspects related to treatment, including changes, according to the individual’s experiences throughout the course of the disease [13]. The explanatory models theory refers to the meaning and sense that each of the individuals involved in the clinical process have about disease and treatment [15]. The explanatory models theory emphasizes the interaction between the patient and health professionals [13].

The illness trajectory theory centres on a series of social and cultural events that have been traditionally interpreted only in medical terms [17]. It works not only on the physiological events, but also on what is intimately related to how an individual defines his/her illness, given that an ill person can define such trajectory differently to relatives and doctors. The illness trajectory theory is a conceptual model that succeeds the idea that chronic diseases follow a variable and modifiable course [17–19]. The following stages conform the illness trajectory theory: perception of symptoms, contingencies, components of the disease, temporal horizons and impact on the support network [17, 19–21].

Patients and methods

This study was based on qualitative field, a design developed by Crabtree and Miller [22] that gathers patients narratives through face-to-face interviews in the clinical setting. Its analysis relied on the interpretative grounded theory methodology and explanatory models [15, 19, 21]. Research process consisted of:

(i) **Data recollection** through in-depth interviews based on a guideline developed by the investigators on the following themes: illness experience, school setting, religion, alternative and complementary medicine, disability experience and patient–physician–relatives relationship. The guideline was flexible and allowed the incorporation of themes as they emerged throughout the study;

(ii) **Coding and analysis** through Charmaz’s interpretative grounded theory method [19, 20] beginning with open coding terms, for example ‘experiencing the diagnosis’;

(iii) **Data interpretation** from the individual and group perspectives; and

(iv) **In reflexivity**, the last part of the process. We followed three theoretical positions to integrate the narratives and the researcher’s interpretation: explanatory models, suffering and illness trajectory [22].

In-depth interviews were carried out and recorded by I.P.-B. in two to four sessions lasting 1–2 h each. Parents were interviewed in the absence of children and vice versa. According to the participation observation technique, complementary notes were taken on the interviewee’s behaviour and context. Themes and questions were adapted to parent’s and children’s characteristics. Tape recordings were transcribed verbatim into a word processor, read for four to six times and imported into the ATLAS\textsuperscript{ti}® 4.2 software.

Analysis followed the constant comparison method [23]. I.P.-B. did the first analysis. Then, analysis and interpretation were triangulated to avoid bias: physicians, psychologists and anthropologists reviewed the data, and met several times to discuss and interpret the results. All themes or codes were initially grouped as family codes and then as concepts, as expressed by the participants. Ultimately, we presented the parents with the results of the study in a meeting.

We included 16 adults and six children from 10 families out of the members of 20 families invited to participate in the study. The group consisted of five males and 11 females (mean age 38±6 yrs); children were four girls and two boys (mean age 13±1 yrs). There were no specific exclusion criteria; children had systemic, oligoarticular or polyarticular JIA differing in disease activity and damage, disease duration, extra-articular manifestations, and past and current treatments. Some patients attended the public hospital and belonged to families where members had been poorly educated and had low incomes while others—seen in the private setting—belonged to educated and wealthy families.

The study was approved by the local institutional review board. Parents and children signed or assented an informed consent, and knew that they were not obligated to talk about themes or give answers to unwanted questions and that they were free to interrupt or even terminate the study at any moment. Parents always selected the site of the interviews.

Results

We identified a major code category resembling the religious reference to a trajectory of pain, faith and hope consistent with the illness trajectory theory [17, 19] that received the name of ‘el peregrinaje’ (pilgrimage) by one of the parents.

‘We went on a pilgrimage with him and the doctors. Everywhere... They couldn’t tell us what was wrong with him.’ (F:85–7).

Pilgrimage was similarly painful for children and parents, but some of the processes and factors differed between them.

**Pilgrimage in children**

Pilgrimage was conformed by immediate concepts, which dealt with a chronic and disabling disease, and on the other hand, with the inability to integrate into peer groups. Children’s lack of knowledge about the origin and future of the disease prevented them from giving any useful meaning to their illness. In consequence, children appeared unable to conceive any treatment as a real cure.

**Pilgrimage in parents**

For parents, pilgrimage was conformed by historical and immediate disease experiences. While parents of systemic JIA children suffered the most during the pre-diagnosis stage, the post-diagnosis stage was linked to healing. Parents of children with oligo/polyarticular JIA appeared to suffer less in the pre-diagnosis stage, but after diagnosis, they had to deal with chronicity, prolonged treatment and little hope for healing. Parents had to integrate JIA in both their individual and social lives.

**Pilgrimage in the pre-diagnosis stage**

While most children’s narratives were poorly informative of JIA onset, some recalled physical pain or functional limitation. Children were mostly informed by their parents—rarely by the physician—on the cause of their discomfort.
Illness trajectories in Mexican children with JIA

1401

Panel started when symptoms appeared. Parents looked for explanations and solutions by whatever means. Initially, symptoms appeared irrelevant to some of them who started looking for help among relatives, friends or ‘sobadores’ (a folk curer). Lack of improvement led to multiple visits to the doctor.

‘...first my mom put me in another [hospital], but they kept telling her I had nothing. And she took me to a lot of hospitals, they said no; that I had nothing wrong’ (J:78–83).

Getting the wrong diagnosis, for example a deadly illness, was worse than getting no diagnosis at all.

‘He was talking to mom, and cried and cried, saying I was going to die and all. And my mom said that if I was meant to die, I would die, but they wouldn’t give it; but that doctor didn’t know’ (R:160–197).

Shortly after onset, children’s and parent’s explanatory models collided with that of the physician. Despite the fact that parents of systemic JIA children knew that no cure will ever be reached, some found relief when JIA was diagnosed. Others mistrusted the physicians’ diagnosis and did not easily accept it as a consequence of wrong diagnoses in the past, interruptions of medical care, and the idea that their children were targets of an experiment. Anguish and despair grew as the result of children’s worsening condition and lack of explanations. Some parents found physicians to be certainly dishonest.

‘Uncertainty is a murderer’ (N.515). ‘They feel they’re unquestionable. I mean, they have God’s truth and no one can doubt it. Even less if you have no medical education. They make it look as an offence if a squealing woman in pain dares question any of their choices.’ (LM:551–554). ‘My daughter’s case was the guinea pig of everyone around her...there we were five more doctors of all ages came along. So they start looking at her and the [head] doctor says that we might be talking about a tropical disease. And I said, you know what, doctor? I might look stupid to you, but I’m not’ (R: 262–280).

Pilgrimage in the post-diagnosis stage

Parents expected some therapeutic improvement to reduce their anguish. The explanatory models and type of care sought by parents depended on their socio-cultural background. While those in the low class looked initially for help among relatives and laypeople, upper class parents first looked for medical care.

Nearly all parents experienced uncertainty and anguish in the first medical consultation and were mostly unsatisfied with medical explanations. Physicians not always allowed questions, particularly when making mistakes. Parents turned to alternative and complementary medicine, but returned back to medicine after finding no relief from the disease. Mothers were often discredited and their observations were doubted by their friends, family and medical staff during the answer-seeking process, despite the fact that they could be the most important source of information.

After diagnosis, pilgrimage went on to more complicated and reflexive steps. Children realized that life would never be the same, and that their physical independence and relationship with other people would radically change.

Explaining the disease

While some parents and children fell into a denial or unfounded hope for a potential cure after certain improvement, others experienced awe, sadness, despair and even guilt. Only a few dealt with disease chronicity as a challenge and a struggle to survive, or with resignation. JIA was experienced so closely that it became a ‘part of the family’.

Explaining JIA to children did not appear to be relevant in their life: they did not understand the physician and seemed not interested in any explanation. Some children knew that they had arthritis, few mentioned ‘juvenile rheumatoid arthritis’, but none knew what exactly it was.

‘...I won’t be doing any girls stuff like washing or jumping and, hub, helping my mom. And that I won’t be normal like everyone else.’ (J: 439–450). ‘You know what I want? I want to be like other kids, play football, basketball...but I can’t’ (L:104–112).

In general, children explained JIA through the words of their parents or the physician. Some children lived their disability when comparing with healthy or other arthritic children.

‘...my mom says that she probably didn’t eat well when she was pregnant. (J:154–166). ‘Well, I feel really bad [when I see other children with JIA]. I think of what’s wrong with them and that I still have much to give’ (I:123–128).

Chronicity

The next stage involved the comprehension of chronicity and physical disability. JIA represented a new disabling condition requiring special care and adjustment of future expectations.

For some children, a wheelchair was a tool against isolation, but in others, a disappointing experience.

‘I was feeling bad because I wanted to go to school and play along with my classmates. But I couldn’t. Because I had no chair and my...hurt so bad and I couldn’t stand...Now that I have the chair, I’m feeling a lot better’ (JM:190–197).

The socio-cultural background influenced the understanding of JIA explanatory models. Parents with a low socio-cultural background knew little about JIA challenges and even hardly recalled its name. Their explanatory models included a mix of parent’s beliefs and physician’s information: disease resulted from the lack of medical care during pregnancy, thermal changes, traumas and even ‘supernatural’ causes. Highly educated people got information from the web, books, doctors and friends and their explanations were greatly loaded with biomedical information, sometimes wrongly interpreted. The socio-cultural background had a major role in decisions regarding medical, alternative or complementary therapy.

‘...I've seen that homeopathic medicine helps better than allopathic. To be honest, I think doctors are forgetting that body, mind and soul are just parts of a whole. They focus on the physical aspects and overlook the rest’ (N:17–20).

In some cases, explanatory models changed over time. For example, a patient who initially attributed the disease to thermal changes and inappropriate care in pregnancy turned to shelter and consolation through religious activities because JIA was a ‘test of God to prove them strong’.

Several circumstances, including mother’s exhaustion, patient-physician relationship, teachers’ unwillingness to sympathize with ill-children and low income, aggravated the disease. Ignorance about JIA—from the biomedical perspective—was common to all socio-economic groups, but the search for information was common among high socio-economic class parents. The more information they got, the closer the patient-physician relationship it was. Regardless of family and individual efforts, people felt that they only prevented further deterioration. Chronicity threatened the meaning of treatment. Family cohesion, support networks and interpersonal relationships deteriorated as a result of an unavoidable existence.
Discussion

The approach of this study was based on the illness trajectory theory, a conceptual model based on the idea that the course of chronic diseases is variable and modifiable throughout time [17, 19, 23]. Through this model, we could explain the trajectory of children with JIA and their parents. JIA was perceived as a pilgrimage, a form of a never-ending process creating a great deal of suffering in children and parents.

Pilgrimage also shared some of the properties of health-seeking [24] and illness behaviour [25, 26] theories to some extent. Health-seeking refers to the sequence of changes that add to a patient’s notions of self and to the system of images that he/she uses to judge himself/herself and other individuals [24]. In contrast to pilgrimage [a historical trajectory triggered and determined by a specific event (disease)], however, health-seeking focuses on the relationship taking place at the institutional level, and overlooks the individual and cultural experiences of illness. Regarding illness behaviour, it focuses on the understanding of chronicity, but disregards the supporting network, spirituality and future expectations [24, 26]. Illness behaviour lacks some of the properties needed to understand suffering and ranks psychological aspects and the utilization of health services above illness experience in the cultural setting [24, 26]. The concept of healer/doctor shopping—the quest for a diagnosis matching the experience of patients to ‘buy the diagnosis’ [27]—was consistent with pre-diagnosis pilgrimage (changes from laypersons to doctors, from one doctor to another, from one institution to another, from medical care to alternative therapies and vice versa). However, pilgrimage aimed for a favourable prognosis and cure, and the diagnosis was sought by the patients, their parents and health professionals.

Five stages of the illness trajectory theory were identified in pilgrimage [17, 19–21]. According to our interpretation, the pre-diagnosis stage of pilgrimage was consistent with the perception of symptoms. For most parents, particularly those with systemic JIA children, receiving a diagnosis was a positive solution, but for some others, it represented a negative solution because chronicity discarded any possibility of cure. On the other hand, children seemed to perceive their diagnosis in a negative way.

Contingencies related to aggravating factors and socio-cultural background. Poverty, migration and family violence were the greatest aggravating factors. Middle and upper-class families perceived negative attitudes, rejection, lack of communication and incompetence while relating with physicians. Components of disease relies on the context in which the patient and his supporting network are set. Parents of systemic JIA children found pre-diagnosis the hardest stage of pilgrimage. For those with oligo/polyarticular JIA children, the worst came when they learnt about the disease itself, its disability and treatment.

Temporal horizons, which in the illness trajectory theory reflects the course of the disease, resemble the changes of disease experiences in the clinical setting [28–30]. Parents and children adjusted and adapted every aspect of life from time to time. The impact of disease on the patient and the supporting network could be associated with the consequences of physical disability: social isolation, loss of independence and harsh relationship with other children. It also influenced the meaning of treatment and any expectation of improvement.

Post-diagnosis pilgrimage spanned from the time of diagnosis to the comprehension of chronicity, physical disability, treatment and future expectations. The first stage was triggered by JIA and began with the so-called disruption in the biography [13, 18]. The second stage—experiencing chronicity—referring to hope or healing expectations was related to psychological aspects, effect of the disease and information. The main concern in JIA was physical disability; thus, whenever parents referred to healing expectations, they actually denied their children’s disability. Interestingly, in cancer patients, the opposite occurs; the greater the healing expectations the lower the level of anxiety and the better the QOL [26]. Parents of JIA children with healing expectations rejected the fact that there was no cure for the disease and were not willing to accept the irreversibility of chronicity [28, 31, 32].

The spouse’s support was a critical issue in this study. Overall, children relied on their mothers as their main support. Parents often stressed that family cohesion and support were keystones to survive the disease, particularly when a nuclear family and other relatives were involved. Interestingly, JIA may in some cases positively affect the network support, particularly when treating the disease and improving children’s QoL. In others, problems increase. While mothers, as caregivers, of children with cancer are encouraged by the disease to a major reorganization of life from work to recreation [32], the mothers of children with JIA who understand chronicity in its full scale may become depressed at the time of diagnosis [28, 31–34]. Notwithstanding, husbands and health professionals tended to disqualify or diminish the role of the mother similarly to behaviours described in families of cancer children [29, 32, 35]. These attitudes appeared to distort and delay the diagnostic process.

Knowledge of disease as well as treatment and preserving the family nucleus are positive strategies in facing the disease [28, 29, 34, 35]. This study identified both strategies: initially, parents faced JIA negatively, but once they accepted the disease their approach turned positive. There were parents, however, who lingered in denial despite the fact that their behaviour could easily fit into the positive strategies.

Physical disability played a major role in children’s narratives by reinforcing the perception of chronicity, diminishing the meaning of treatment and leading to social isolation. Lack of improvement proved the insignificance of the treatment and interactions with healthy children made them realize their physical disability. Time-consuming treatments, dependency and discriminatory attitudes of caregivers led the JIA children to increased isolation [28, 31, 36].

In contrast to previous reports [34, 37], children with JIA showed a positive attitude—reflecting great acceptance of disease and working against isolation—when using certain aid devices. This phenomenon, described as the ‘disability paradox’ [36], may have two different forms: the first one refers to people with daily-life serious limitations experiencing social discrimination, but carrying on with their lives normally; and the second to health professionals and people who usually label the QoL of disabled individuals as poor [17, 36, 38]. As previously described [28, 31, 33, 37, 39, 40], low treatment compliance was a major issue in our study. However, parents and children know that interacting outside the hospital with children in the same circumstances is a key aspect in building treatment compliance [28, 31, 37, 39, 40].

Study limitations

Verbal communication is prone to distortions, exaggerations and even false data. Some of the things that people say and do depend on the circumstances; an interview is not real life. Since the interviewer does not observe the individual in his daily-life activities, he is not able to appreciate the context in which events take place and might not understand the whole of the patient’s perspective. Due to the fact that we could not interview some parents and children, our research might be limited; however, we did not expect the appearance of themes not previously identified. It seems that in children some experiences were genre related, yet unfortunately we could not interview some more boys and girls to confirm such an impression. Despite the fact that the scope of this study did not include physicians involved in JIA children’s care, it seems necessary to get the narratives of their experiences and complement the illness trajectory of these patients. The fact that this study was carried out in a specific cultural group does not limit the scope of this
study since Mexican culture is shared by other Hispanic countries, and a significant proportion of the population of developed countries is actually Hispanic.

In conclusion, pilgrimage, conceived as the process of living with a chronic disease such as JIA, is perceived by children and their parents in different ways. Their concerns and the impact they suffer stand at different levels. Parent’s narratives as proxy do not replace those of children, but they work as mutual complement. According to this study, reconstructing the illness trajectory of JIA in context of individual and general explanatory models would allow us to identify a number of cultural issues involved in children and parent suffering. With this type of information at hand, it is expected that the patient–physician relationship would improve, and children and parents would receive a much more appropriate medical care.

The authors have declared no conflicts of interest.

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