Contribution of the use of basic telemedicine tools to the care of children and adolescents with juvenile idiopathic arthritis at the Puerto Montt Hospital, Chile

Aporte del uso de herramientas básicas de Telemedicina en la atención de niños y adolescentes con Artritis idiopática juvenil, en el Hospital de Puerto Montt. Chile

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Abstract

Children and adolescents with rheumatologic diseases require specialized and comprehensive care, but pediatric rheumatologists and immunologists are concentrated in hospitals with specific, high-cost and modern technology. Considering that some patients with juvenile idiopathic arthritis (JIA) live in rural, remote and limited accessibility areas, the use of Telemdecine (TM) can optimize diagnosis, follow-up and prognosis. **Objective:** Reporting 10 years of experience of a mixed care model: face-to-face and distance, using basic TM; the institutional impact, advantages, disadvantages and acceptance informed by parents and patients. **Patients and Method:** Exploratory, descriptive, and retrospective study with qualitative component. After the authorization of a scientific-ethics committee of the Reloncaví Health Service and the application of informed consent, a review of medical records was carried out and a qualitative survey was applied to parents and children over 14 years of age with JIA, seen between 2005-2015 in the pediatric ambulatory rheumatology polyclinic of Puerto Montt Hospital. **Results:** The were 27/35 participating patients with JIA attended by a trained pediatrician and assisted by distance (1,000 km) by an immunologist. The 8/35 patients did not answer by choice or change of address. The 70% of parents and patients accepted the model of care and 4% would prefer sporadic care only by specialists for diagnosis and follow-up. The number of patients transferred annually decreased from 10 to 1. The advantages of the care model outweighed the disadvantages perceived by parents and JIA patients. **Conclusion:** The use of TM tools in JIA decreased transfers, improved follow-up and were considered advantageous by patients and their parents.

Keywords:
JIA, quality health care, telemedicine, pediatric immunology-rheumatology, care model

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Introduction

In 2010, WHO defined Telemedicine (TM) as “The delivery of healthcare services, where distance is a critical factor, by all health care professionals using information and communication technologies for the exchange of valid information for diagnosis, treatment and prevention of disease and injuries, research and evaluation, and for the continuing education of healthcare providers, all in the interests of advancing the health of individuals and their communities”1. Models of care that use TM have the potential to at least partially correct the poor geographical distribution of specialists and reduce inequalities in the quality of care provided to children with chronic or complex pathologies living in rural and/or remote areas where there are no specialists2. Juvenile idiopathic arthritis (JIA) defined by the International League of Associations for Rheumatology (ILAR) as arthritis of unknown etiology, which begins before the age of 16 and lasts for at least six weeks, excluding other known conditions3-4, is a disease covered by the system of Explicit Health Guarantees (GES) of the Ministry of Health of Chile since 2010, which ensures diagnostic confirmation by a specialist in less than 30 days and specialized, multidisciplinary and comprehensive treatment, including periodic follow-up and rehabilitation5. The JIA often persists into adulthood and can lead to morbidity and physical disability, particularly in cases of late diagnosis and/or suboptimal treatment5-6. In Chile there is a deficit of pediatric immunologists and rheumatologists, who are concentrated in Santiago, the capital city where the national reference centers are located 1,000 km away from Puerto Montt7. In 2005, a progressive model of mixed care was implemented, with initial evaluation by a pediatric immunologist in Santiago de Chile, and follow-up in Puerto Montt by a pediatrician trained in JIA, with immunological advice via telephone, e-mail or written/text message. The objective of this study is to show ten years of experience of a mixed model of care: face-to-face and at distance, using basic TM; its institutional impact, advantages, disadvantages, and acceptance reported by parents and patients.

Patients and Method

Descriptive retrospective study. With prior informed consent/acceptance, the clinical records of all patients with suspected JIA controlled at Puerto Montt Hospital (HPM) were reviewed from January 1, 2005, to December 31, 2015. The variables considered were: age, gender, place of residence, distance and duration of the journey to the HPM, year, place and doctor who made the initial diagnosis, age of diagnosis and start of treatment, type of JIA, use of biological treatments, follow-up and frequency of direct attention by a specialist in Santiago and by a trained pediatrician at HPM with remote specialist advice (e-mail, telephone, written/text message), and number of patients transferred annually to referral centers. Physicians and pediatric nurse, who did not participate in clinical care, sent and collected a self-administered qualitative survey to parents and children over 14 years of age regarding the model of care in Puerto Montt, by a pediatrician trained in JIA, who was advised remotely by specialists. For statistical analysis, Mac version STATA 13 software was used.

Inclusion Criteria

Children and adolescents under 16 years old, with a confirmed diagnosis of JIA in national reference centers, according to ILAR criteria, seen by a pediatrician trained in the polyclinic of “pediatric rheumatology” of the HPM.

Exclusion Criteria

Patients with JIA suspicion and definitive diagnosis of another pathology. Patients diagnosed with JIA and treated exclusively in Puerto Montt.

Results

Out of 50 patients with suspected JIA, 42 corresponded to JIA according to ILAR criteria. 8 patients had other pathologies: 2 patients with acute leukemia, and 1 patient with each of the following pathologies respectively: villonodular synovitis of knee, arthritis due to Bartonella, parvovirus arthritis, dorsolumbar myelomeningocele sequel, fibromyalgia and conversion disorder. Out of the patients with confirmed JIA, 83% participated in the mixed care model. Figure 1 shows the clinical and demographic characteristics of all subjects. Currently, there are no patients under 4 years of age in control, 17 patients are over 16 years of age, 14 adolescents have been transferred to adult rheumatology, and 3 are in transition (figure 1A). Regarding the age of diagnosis, 12 patients were diagnosed before 4 years of age, 2 of them before 2 years of age, and some patients had late diagnosis, consulting before 16 years of age but later confirmed, considered as JIA in relation to the onset of symptoms (figure 1B). Consistent with national and international data, women predominate in a relation of 3:1. The average age of diagnosis is 8 years 2 months and the period between onset of symptoms, diagnosis confirmation and start of treatment is 1 year 5 months. The distribution by JIA subtype is: 33% polyarticular with RF (-), 26% oligoarticular (21% pure oligoarticular and 5% exten-
ded oligoarticular), 17% polyarticular RF (+), 12% systemic, 7% psoriatic and 5% associated with enthesitis (figure 1C). Regarding treatment (GES incorporates and guarantees access to biological medicines since 2013), 48% of the patients are taking biological medicines (figure 1D), due to intolerance or lack of response to methotrexate, used as first-line treatment in all patients, suspended or associated with biological medicines; etanercept, adalimumab, infliximab, abatacept and tocilizumab, sequentially, according to the established response criteria5,8-12. Figure 2 shows the distances in kilometers and travel time, from the residence to the Puerto Montt Hospital, 73.1 km average region [min. 1, max. 202] (figure 2A) and 1.4 hours average region [min. 20 min, max. 4 hours] (figure 2B) demonstrating the distance and travel time required to reach the HPM, which could add up to 1,000 km to the centers of reference. Figure 2C shows the incidence variations from 1998 to 2015, with the highest number of cases diagnosed in 2015, whose cause has not been identified. Finally, the 2D figure shows the decrease in travels for diagnosis and follow-up from Puerto Montt to national reference centers (patients diagnosed between 1998 and 2004 represent 54% of travels; older patients traveled more than newer, patients [The value of r (correlation coefficient) is 0.74; p-value of 0.00].

With respect to connectivity, 100% has mobile telephony and 96% has internet and e-mail access. 27/35 patients under a mixed care model answered the survey (77% of coverage).

Table 1 shows disease awareness and opinions about the model of care: 70% recognizes that it is a joint disease, 70% accepts and prefers the model of care used, and only 1 patient would opt for care only by specialist, while 26% would agree to be treated only in Puerto Montt with remote advice by specialist. The

Figure 1. Distribution according to demographic and clinical characteristics of 42 AIJ patients. A: Distribution according to age until January 2017. B: Distribution according to age at diagnosis. C: Distribution according to ILAR category. D: Distribution according to biological treatment.
spontaneously indicated advantages are better access to consultation, avoid travel costs and times (physical wear and stress-school and work absenteeism) and trust in the treating physician. Although they mostly accept the care model (67-70%), five patients (19%) believe that there could be some risk due to a possible discrepancy between the evaluation of the specialist and the local doctor.

**Discussion**

Patients with JIA treated at Puerto Montt Hospital (HPM), one of the two high-complexity hospitals in the X Region, reside in the provinces of Llanquihue, Chiloe, and Palena, a geographical area of high population dispersion, distributed in island and continental territory of limited accessibility (Google maps). The HPM is 1,000 km away from the reference centers for pediatric immunological and rheumatological pathologies. Before 2005, patients with suspected JIA had to confirm it, treat it and have a follow-up on an outpatient basis, depending on the availability of attention hours at San Borja Arriaran Hospital (HSBA) or Luis Calvo Mackenna Hospital (HLCM) in Santiago, traveling by land for 12-13 hours. In 2005, the first patient is counter-referred from the HLCM to carry out follow-up in the HPM, decreasing the frequency of controls in Santiago, successively adding patients already diagnosed and new ones with confirmed diagnostic
The reasons for the predominance of the polyarticular form RF (-) are not clear, and although there was German colonization in this region, there is no history of patients with German ancestors in our cohort. On the other hand, it is possible that there may exist subnotification of the oligoarticular form, later diagnoses, preferably consulting the forms with higher joint involvement, a probable hypothesis considering the time between the beginning of symptoms and diagnosis (figure 1B)18-26. Also noteworthy is the distance from home to the HPM, expressed in kilometers and transport times. The concept of TM (healing at a distance) (WHO 2010 ref. 1) or telehealth emerged in the 1970s, with the development of technology (personal computers, internet, and mobile communication devices). The most recent revision corresponds to Dorsey27.

TM applications are multiple, whether real-time or delayed. It has advantages and disadvantages that depend on the time and place of implementation28. It has been applied to multiple acute and chronic clinical
conditions with good results, including those requiring specialist consultation.

It has been used as an alternative or in addition to the usual care (face-to-face consultation or telephone consultation), partially replacing the usual care. TM technology can be grouped into three categories: remote monitoring, data storage and retransmission, and interactive. Another related concept is the exchange of health information, defined as “the act of sharing clinical information between healthcare professionals and places where health care is practiced that are not part of the same organization.”

The chosen model of care will depend on organizational factors and clinical need. In our case, we use basic means (mobile telephone, written/text message, and e-mail), which could be considered in the second category (storage and retransmission of data), since in our health region, interactive telemedicine has not been implemented to more specialized centers, but exclusively from the regional health service to the network of dependent health centers, a situation observed in other countries. In the USA, Italy and the United Kingdom, the use of health information technology (HIT) has been proposed and used in healthcare, covering different chronic diseases, including JIA, recording inequities in the quality of care, its causes and consequences, becoming a challenge to facilitate access to pediatric rheumatology in order to improve prognosis.

Particularly, Consolaro publishes the use of HIT by applying surveys or questionnaires to parents and patients with JIA. Thus, this is, to our knowledge, the first evaluation of a mixed model of care; face-to-face by a pediatrician trained in the management of children and adolescents with JIA, and at a distance by a specialist, using basic means of communication, which could be considered within the concept of Telemedicine, although it differs from the traditional models, in which there is no direct participation of the patient (or his or her responsible relative) with the specialist, but indirect participation through a trained pediatrician, who follows clinical guidelines and requests advice according to clinical necessity.

It has been reported that improving the health status and quality of life of patients and their families are key results in contemporary health care, especially in vulnerable individuals where psychosocial problems are prevalent; therefore, the application of these or other TM tools in places with limited access to specialists could positively impact the overall health prognosis of patients with JIA.

The advantages of the model of care pointed out by parents and patients coincide with those reported using TM: A reduction in patient and family transfers, and absenteeism from school, work and home; decreased uncertainty and worsening of pain associated with inflammation and prolonged immobility (12 hours of travel), in addition to reduced personal food costs. An increase in the number of check-ups per doctor, a higher ease in dispensing medication, ophthalmologic evaluation, rehabilitation unit assistance and therapy changes, including early access to biological therapy. Knowledge of the disease improved, allowing increased adherence to medication (table 1).

Within the regional health organization, the institutional cost associated with transportation also decreased. Other benefits not shown in this study are nursing and medical training (assessment of patients, learning, and reduction of stress involved in the management of complex patients, application and monitoring of methotrexate and biological drug use,) networked to health centers near home. The outcomes of this study have methodological limitations and should be interpreted with caution. First, the model was introduced progressively and without prior planning, and could thus be considered an exploratory study. Consequently, we are not looking for statistical significance, but rather for clinical differences. Secondly, this experience was carried out in a single center with particular geographical, demographic and health resource characteristics, which limits the generalization to the entire population of patients with JIA, although it could be replicable in centers with similar characteristics.

Our goal was to evaluate some results of a 10-year application of this model of care, which could be planned and perfected by using more modern tools, according to the current characteristics of the population and the technological means available, incorporating more actively the patient and his or her family.

At the same time, it highlights the urgent need for the training of specialists within the public health network and their distribution in regions of the country furthest from the reference center, a common situation in developed countries as well.

There is no doubt that better designed, controlled and randomized trials are needed to draw more scientifically powered conclusions about the effectiveness of this model of care.

**Ethical responsibilities**

**Human Beings and animals protection:** Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

**Data confidentiality:** The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.
Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

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