Life for patients with myelofibrosis: the physical, emotional and financial impact, collected using narrative medicine—Results from the Italian ‘Back to Life’ project

The living of patients with myelofibrosis: physical, emotional and economic impact of the disease collected through Narrative Medicine

The results of the Italian project named “Back to Life”

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Abstract

PURPOSE: Myelofibrosis (MF) is a chronic myeloproliferative neoplasm clinically characterized by cytopenias, progressive splenomegaly, invalidating constitutional symptoms and reduced survival. While patients experience a severely impaired quality of life, their families face the practical and emotional upheaval of the routine of everyday life together with the issue of disease-related financial costs. The present study aims to investigate how patients and their caregivers perceive their living with MF in Italy, and their Burden Of Illness.

PATIENTS AND METHODS:

A quali-quantitative semi-structured questionnaire and a narrative plot were administered to patients affected by Primary or post-Essential Thrombocythemia/post-Polycythemia Vera MF and to their caregivers in 35 Italian haematological centers. Patients and caregivers were enrolled on a voluntary basis between September 2012 and October 2013.

RESULTS:

287 questionnaires from patients and 98 from their caregivers were collected; additionally, 277 narratives were obtained, respectively 215 from patients and 62 from caregivers. At diagnosis communication, the most patients' expressed emotional states were fear, sorrow and anger, confirming the difficulty in living this phase. A high level of emotional distress was found also in caregivers. Along the pathway of care, the ability to cope with the disease was different according to the quality of care received. Furthermore, the coping strategies were more developed among patients with a good caregiving within families and satisfying relationships of care with physicians. Costs attributable to MF were estimated: 12.466 € per year from the patients' perspective, the average annual cost of loss of income was assessed at 7.774 € per patient, 4.692 € per caregiver.

CONCLUSIONS:

The understanding of the personal life of MF patients and their families may be crucial to improve the human relationships between health workers and patients, to build a more targeted organization of the medical pathway of care and to drive a more effective financial support to allow the patients to maintain their productivity and social role.

Introduction

Primary myelofibrosis (PMF) is the most severe of the Philadelphia-negative chronic myeloproliferative neoplasms (MPNs) and is characterized by expansion of clonal hematopoietic cells with release of cytokines [1,2,3]. Usually, MF presents with cytopenias, leukoerythroblastosis, extramedullary hematopoiesis, and progressive splenomegaly that may be accompanied by hepatomegaly [4,5]. MF can also arise from other MPNs, i.e. polycythemia vera and essential thrombocythemia (post-PV or post-ET MF) [6]. Discovery of the V617F mutation of the Janus kinase (JAK) 2 gene, and more recently of MPL and CALR gene, represented an important step in the understanding of the pathogenesis of MF. However, the genetic trigger of MF is still unknown [7].

PMF is a rare disorder with an estimated annual incidence of approximately 1 per 100,000 and a prevalence of 4–6 per 100,000 persons in Europe [8]. It affects primarily older individuals with a median age of 67 years, when subjects may be still in their productive phase [9]. Prognosis in PMF can be stratified according to several indexes and median survival is different among risk categories, ranging from 1.5 to 14.2 years [10], but it may extend to over 20 years in low-risk patients [11].

Myelofibrosis not only shortens survival but also severely compromises overall quality of life (QoL) as a result of the profound constitutional symptoms. The Myeloproliferative Neoplasm Symptom Assessment Form (MPN-SAF), which is a validated quality of life (QoL) instrument, revealed a severe symptomatic burden, with fatigue, reduced QoL and ability to work or participate in desired activities, reported in the majority of patients [12,13]. Symptoms experienced by patients are heterogeneous including excessive and disabling fatigue, early satiety, anorexia, pruritus, bone pain, night sweats, cachexia, abdominal pain and discomfort, and cognitive complaints. The only curative treatment is allogeneic haematopoietic stem cell transplantation, a procedure still reserved to a minority of patients [14]. Therapeutic approaches preferably rely on clinical needs and the consideration of IPSS risk category and patient's age and comorbidities. Main conventional treatments include cytoreductive therapies, erythropoiesis-stimulating agents, and steroids. Recently, the JAK1/2 inhibitor ruxolitinib (Jakavi/Jakafi) has demonstrated remarkable success in alleviating MF symptomatic burden, reducing splenomegaly and improving quality of life while offering overall survival benefit [15, 16].

To-date, no studies have been performed to understand the experience of living with MF, that include not only the medical burden of the disease (i.e.: MF-related symptoms, treatments' toxicities) but also financial issues (i.e.: disease-related out-of-pocket costs). While the issue of indirect costs of chronic disease has been addressed in a number of studies, most papers focus on differences among pharmacological treatments, potential savings, lack of productivity and consumption from a public health perspective. Yet the indirect costs from the patient's perspective

— more complex to identify and quantify— are still largely underestimated. Indeed, the concept of burden of disease mainly focuses on direct medical costs (e.g. costs for diagnostic tests, therapies and/or rehabilitation) paid by the public health care service or insurances. The extended concept of “economic burden of illness” refers to a broader array of intangible non-medical costs (e.g. transportation costs, household utilities and adjustments, domestic care assistance), indirect costs (e.g. loss of productivity), as well as the caregivers’ incurred costs and their loss of productivity, and lastly psychosocial or intangible costs due to the deterioration in the quality of life [17,18].

This survey reports the results of a quali-quantitative semi-structured questionnaire integrated with a narrative plot aimed to investigate patients’ and caregivers’ perception of living with MF in Italy and their economic burden of illness.

Patients and methods

The study was addressed to patients affected by Primary or post-Essential Thrombocythemia/post-Polycythemia Vera MF (independently from the severity of the disease and the type of treatment) and their family caregiver (defined as the person putting most effort in assistance to patient). The quali-quantitative questionnaire was integrated with a Narrative Medicine approach: a semi-structured questionnaire and a narrative plot were distributed in 35 Italian hematological centers, under the methodological supervision of ISTUD Foundation, between September 2012 and November 2012 and between June 2013 and October 2013. The target of the study was to collect interviews from at least 250 patients and 100 caregivers, so thus to collect 10 testimonies (patients + caregivers) for center of care and obtain a geographically homogeneous Italian repliers' group. Statistical sampling was not the aim of the survey. Interviews were proposed on a voluntary basis, only after releasing written informed consent according to the Italian Privacy Law [19].

The questionnaire covered six sections evaluating different areas of illness impact: (1) socio-demographic information, (2) clinical status at diagnosis, (3) clinical status after treatments, (4) implications on the working productivity (before and after the disease diagnosis), (5) out of pocket costs, (6) caregiving assistance. Caregivers' questionnaire included the Caregiver Self-Assessment Questionnaire to measure the stress level linked to the caring for a patient with a chronic disease [20]; this session of the survey was translated from English to Italian without changing any item. [ATTACHMENTS 1 AND 2].

Considering the process of the story of illness, the narrative plot was structured according to Greenhalgh's methodology [21], which identifies three main phases: "falling ill", "being ill", "getting better" or "getting worse". Narrative, in fact, follows a determined plot that begins before diagnosis and continues through the clinical encounter, investigations, development of a management plan, treatment and resolution or progression of the disease.

Data from questionnaires were analyzed using standard statistical evaluation tools, in order to provide descriptive statistics as mean ranges and frequencies. No inferential analysis was performed with any sample size calculation because of the narrative nature of the study. Analysis on specific therapies were not an objective of the present study, therefore information regarding treatments have been analyzed and reported in an aggregated form.

Studying the illness through Narrative Medicine

Narrative Medicine can be defined as "*what is circumscribed between the physician and the patient, from the collection of information on events before the disease focusing on psychological, social and ontological implications"* [22]. Collecting narratives from patients, their families, healthcare and social professionals, allows clinicians to pay attention to how the person lives his/her illness, and faces the problem together with the health care team [23].

The difference between the two words “disease” and “illness” reflects the integration between Evidence-Based Medicine and Narrative Medicine: “disease” describes the event as an alteration of the biological structure or functioning; the word “illness” describes how the person lives with a disease, including the person’s perceptions, feelings and thoughts. Dealing with the “illness” experience is mainly entrusted on the personal sensitivity and expertise of the physician [24, 25].

Narrative allows the possibility to understand patient’s and physician’s needs, which cannot be captured by questionnaires or any other quantitative approach [26].

Patients and caregivers’ emotional spheres and mindsets were analyzed to understand their coping strategies and integrated with the analysis of the Burden Of Illness.

Coping is defined as the conscious effort to solve personal and interpersonal problems, and to master, minimize or tolerate stress or conflict [27]. It represents the pattern of the individual’s inner resources in a stressful situation like a disease; beyond personality, also social factors and the daily context have been proved to be responsible for better coping. Narrative can evaluate the efficacy of coping strategies, revealing their possible presence or absence in living with the disease. There are specific coping activating or deactivating factors: optimism, curiosity, awareness, kindness and responsibility are considered activating factors; obsessive thinking, introversion, negation are deactivating factors [28].

The Burden Of Illness (BOI) is the global load of a disease not only related to the patient but also to his/her family and community of reference. BOI examines physical, psychological, relational, economics, working and spiritual components, which can depend on the disease and its possible side effects and complications. This kind of analysis allows to value direct and not direct costs which can impact patients and their families quality of life [29, 30].

Estimate of caregiving costs

The assistance of the informal caregiver was examined considering her/his accomplished tasks and the time spent in such chores/activities. This information has been quantified throughout the attribution of a specific economic value: the number of hours of assistance provided based on the Italian National Labour Contract for carers in Italy. According to this contract, the cost per hour is established as 4,41€ (“Level A” – generic carer with basic training; cleaner; laundry operator; housework aid) or 7,83 € (“Level Ds”: assistant to not self-sufficient patients, holding a professional degree or a specific certificate – e.g. general registered nurse, geriatric assistant - who also performs tasks related to feeding and house cleaning). When the carer was involved for over 5 hours per day, the monthly cost was considered corresponding to 606.79 € for level A and 1158.42 € for level Ds.

Analysis of illness narratives

Narratives were collected in complete anonymity and analyzed with two different approaches. All narratives were read independently by three researchers to identify the major patterns of living with MF and the coping strategies. The coping aspects evaluated in the analysis of the stories were: openness, agreeableness, extraversion, optimism, consciousness (engagement) or obsessive thoughts, isolation (disengagement) [31]. In order to compare the coping status with the loss of annual income, the stories were categorized into two groups: "stories in evolution", in which the narration featured engagement coping strategies and led to living the disease in a positive way, and "idle stories", in which the narratives showed only disengagement coping strategies.

Together with qualitative analysis, the texts were analyzed throughout an elaboration of recurring words and semantic expressions with the use of the Nvivo10 software, which facilitates a rigorous textual analysis. The software is based on the approach of the Grounded Theory [32], which assumes to carry out the analysis without any previous hypothesis. This tool facilitates theory building from the data, allowing to generate substantive codes: words are clustered in nodes based on specific text search; information can be analyzed through word frequency, comparisons and logical correlations to create models, graphs or charts [33]. Data can be reconstructed to reflect a view of reality.

Results

Patients and caregivers characteristics

Overall, 287 questionnaires from patients with MF and 98 from their caregivers were collected, together with 215 narratives from patients and 62 from caregivers.

The median age of the enrolled patients was 65 years (range 27-89), prevalence of male patients (55%), 73% married. 80% of the patients had children that were still economically dependent in 23% of cases. 38% of patients were already retired at the time of diagnosis. Caregivers were younger than patients (median age 54 years, range 22-86), prevalently female subjects (56%), married (80%), representing in 65% of cases the spouse of the patient. Both patients and caregivers' socio-demographic features showed a majority of secondary school diploma (34% patients and 41% caregivers), similar to that reported in the general Italian population [34]. 44% of patients were affected by PMF, whereas 21% by a post-TE and 19% a post-PV diagnosis. 16% of patients did not provide the answer related to the specific pathology [Tab.1].

The beginning of the pathway: diagnosis and clinical status

In 40% of cases, diagnosis of MF occurred in an unexpected moment, during an everyday context, and it was perceived as an imbalance of the body integrity. In other 19% of cases, patients realized the disease during a course of treatment for other health conditions. The remaining 41% of patients made a specific connection with their perceived symptoms at the moment of the diagnosis and among them 10% stated that they were in good health.

Quantitative analysis showed that diagnosis was made after an average time of 2,5 years after the appearance of the first symptom, and required more than 3 years in 21% of cases. During the generally long period to get the MF diagnosis, 16% of patients asked for 3 or more consultancies: narrative analysis showed the discomfort in the pre-diagnosis period due to this long-lasting time of uncertainty.

The most frequently reported symptom at the time of diagnosis was fatigue (70%), considered extremely disabling by patients as it adversely conditioned their daily activities and forced them to spend several hours in bed during the day. In second instance, patients experienced difficulties in movement caused by splenomegaly (68%), which forced patients to change their habits and limited their mobility, overall causing a strong impact on the patients' quality of life, even in absence of abdominal pain. Systemic symptoms (insomnia, bone pain, itching, loss of appetite, stress, muscle aches) occurred in 63% of patients, while constitutional symptoms (sweating, weight loss, fever) were reported by 49% of them.

A decrease in daily activities was experienced by 52% of the patients during the course of the

disease (31% due to the increased splenomegaly): in particular, 49% reported limitation of outdoor activities, 23% of domestic activities, and 6% of self-daily ability activities.
Respondents were asked to evaluate their quality of life before starting treatments: it was reported as excellent in 8%, good in 32%, tolerable in 31%, poor in 20%, very poor in 4% and very bad in 5% of cases.

The communication of the diagnosis

The most critical moment reported in the relationship between doctor and patient was the communication of diagnosis, when hematologists explained MF as a chronic cancer with no standardized therapeutic management. The most frequently remembered words by patients in the first description of this disease were "*chronic disease*", "*rare pathology to be monitored*", and *"curable leukemia*". Indeed, the most common word used by patients with secondary MF was "*evolve*", as PV or ET transformed to MF. Therefore, medical language, although precise and accurate, was perceived as too technical and subject to misinterpretations. In most cases, patients reported that the setting and time dedicated to explaining and commenting the diagnosis were not adequate and did not favor a good understanding and a satisfactory emotional acceptance of the disease.

Qualitative narrative analysis reported that the most frequent emotions felt by patients, at the time of diagnosis communication, were fear (53%) and sorrow (23%), followed by relief (11%), resignation (7%) and anger (6%). Moreover, the narratives highlighted the inconvenience of not having initially completely understood the disease; the patients felt ignorant and uninformed: "*I felt lost since I didn't know the disease and what would be its evolution*"; "*I felt worried since I hadn't heard talking about this disease before*"; "*I didn't know anything about the disease but the conversation with Doctor P. got me worried*". After the first moment of diagnosis, 47% of patients continued to express resignation and despair; these people felt helpless and in some cases they decided not to react, just waiting for the death. On the contrary, 27% of narratives showed the patients' inner strength: this group of people decided to react and find out a way to live with the disease. Still, 6% of patients tried to be strong for their family and 18% thought more pragmatically to the difficulties to face for their ordinary life [Tab. 2].

Patients' life with myelofibrosis along the pathway of care

Before beginning a therapy, 52% of patients stated not to be able to perform their normal daily activities anymore. The most impacted activities were walking and travelling (49%), or domestic activities as cooking and housekeeping (23%). MF impaired the essential and daily activities as dressing, washing and feeding, in 6% of cases.

In all narratives, therapy had a pivotal role in patients' experiences, defined as useful and effective in most cases. As a consequence of therapy, the majority of patients reported improvements of signs and symptoms associated with the disease, such as splenomegaly (43%), systemic symptoms (58%), and constitutional symptoms (61%); these improvements were also described in 91% of narratives. On the contrary, 29% of patients did not report any improvement [Table 3].

48% of respondents indicated an improving in their quality of life and 52% of them restarted their main daily activities previously interrupted due to MF-related clinical problems [Tab. 4]. 60% of patients were able to maintain their working activities, although 20% of them reduced their productivity and in other 20% of cases they had to interrupt their professional life. The house-keeping activity is demanded to other helpers in 10% of cases, even after treatment.

From questionnaires, therapies appear to improve patients' moods and reduce fear (54%) and depression (36%). Also, narratives reported that the initial MF patients' feelings of fear and pain gradually diminished over time, due to the health improvements, a better knowledge of the disease and an acquired familiarity with the medical center of reference.

At the beginning of the disease, only 35% of patients declared to be able to live peaceful relationships with their relatives and to speak openly about their condition. The remaining patients reported that their relationships were negatively influenced by emotions of fear, anger and grief; in some cases, problems of isolation were evident. Relatives and friends close to the patients were often described as worried but unable to be a point of reference and support. After the Hematology center took charge of the patient, the family situation seemed to improve, and the proportion of patients living serene relationships with their family members increased to 60%. At the end of the narrative, the future was expected full of hope and courage in 70% of the narratives, even if almost half of the patients stated to hope for a complete recovery, not accepting MF as a chronic disease.

Life with myelofibrosis: the caregiver's perspective

Caregiving was carried out within families: 65% of the respondent caregivers were represented by patients' spouses (wife 37%, husband 28%), while the remaining 35% was composed by other closed family members (daughter 18%, son 11%, brother 3%, sister 2%, nephew 1%) [Figure 1]. The duration of the caregiving was longer than 4 years in 52% of the cases, between 2 and 4 years in 20% and between 1 and 2 years in 14% of the cases; in 55% of the cases, the respondents were assisting people in good general health conditions and able to preserve their autonomy. Similarly to the patients, the most difficult moment faced by caregivers was the communication of the diagnosis: the most frequent emotions were fear (28%), discomfort (28%) and bewilderment (18%). Despite these negative feelings, 45% of caregivers declared an immediate desire to find the strength to react and stay close to their loved one, while 23% of them decided to look for the best treatment.

The "Caregiver self-assessment questionnaire" revealed a very high level of stress in 87% of the cases; as evidenced by 40% of the narratives, caregivers suffer for the patients' fear (23%), anger (19%) and sorrow (10%), emotions which make more difficult their task, as confirmed in their narratives: "*I feel misunderstood and abandoned*"; "*I see my husband suffering and I feel powerless and not prepared to assist him*"; "*I often cry*"; "*I tend to stay isolated to avoid to be a burden*".

In most cases (70%), caregivers reported that the disease had a negative effect on patients, but they still decided from the beginning to be closed to their relatives with MF (44%). All caregivers felt to be taken into consideration by medical doctors.

Thinking about the future, feelings such as fear, anxiety, and sadness emerged in 37% of the caregivers' narratives, while positive feelings of confidence, serenity and optimism, occurred in 27%. The remaining 36% of narratives revealed hope as a predominant feeling: the hope of a "miracle" of healing, wishing for a divine help or the discovery of a new treatment – revealing the same lack of acceptance of the chronic disease, as well as patients -; or the hope to live peacefully and exploiting every possible moment with their loved ones.

Economic impact of MF for patients and caregivers

91% of patients reported to refer to the Hematology Center located close to their homes, in their own Region of residency. The average travel time to the hospital was 55 minutes (range: 3-720). The majority (87%) of responders usually went to the hospital by car, while 7% used train or bus (3%). The average transportation cost corresponded to 42€ per visit (range: 2-600). Direct costs to access the healthcare system resources (ticket, examinations and tests) were reported by 12% of patients (in average 137 €/month and 1.644 €/year).

At the time of the interview, 86 out of 144 patients (60%) had not stopped working nor reduced their work activity after MF diagnosis, and were still maintaining their usual working activity. As a consequence of the disease, 63 patients (22%) reported an annual earning loss of 7.774 €. Overall, 63% of patients needed daily assistance of an informal caregiver. Requested activities were household duties (49%) and chores outside the home (51%). Also, caregivers provided social support, which mostly consisted in not leaving the patient alone. Overall, 41% of caregivers spent more than 6 hours/day for this activity. The estimated monthly average cost corresponding to the informal caregiving was 655 €, which resulted in a yearly value of 7.860 €, ranging from 6.564 up to 8.796 €/year. Additionally, caregivers reported a reduction in their annual earning after MF diagnosis, for an average annual earning loss of 4.692 €. [Table 5]

The analysis of coping strategies from patients' and caregivers' narratives

Referring to Launer and Robinson’s classification of narratives [35], testimonies can be divided into “progressive” and "stable" stories. Progressive narratives showed the individual’s ability to cope even under a difficult and uncertain situation. Conversely, in “stable” narratives the person did not find the way to efficiently manage the situation and her/his situation remained chaotic and tangled over time.

Patients’ narratives were classified as “progressive” in 48% of cases. The factors that allowed the positive development of their stories were, primarily, the recovery, or maintenance, of the daily activities and the success of therapy. In addition to the clinical improvement of the disease, other coping key elements were the opportunity to share, debate and get support with/by others. Family members were able to provide the needed support, helping patients to understand that a satisfactory life was still possible. Furthermore, the relationship of trust and support with physicians allowed patients to understand the course of the pathology and made them feel aware, reassured and cared for. As a result, patients trusted the treatment, considering it the best possible solution.

On the contrary, patients who wrote stable narratives described their physician as professionally and technically prepared, but less empathic and helpful (only in 22% of cases they were satisfied about them) [Tab. 6 and 7].

51% of caregivers’ narrations were classified as progressive, since the strong and motivated dedication to their loved ones was the positive stimulus to make the narrative “evolving”. Two categories of feelings were detected: the first one was a “moral” sense of otherness, respect for the family and willingness to continue, to struggle and fight together, regardless of the outcome of treatment. The second category was related to the sphere of affection, where the word “*love”* was one of the most frequent. Love enabled caregivers to endure the constant mood swings and the moments of difficult relationships,.

The type of evolution of the narratives was found to be correlated to cost issues. Patients who wrote a “progressive story” were those also reporting lower indirect costs, being able to save up to 1.365 € (-18%) compared with the average annual loss.

Discussion

Overall, the integration of Narrative Medicine approach with a quali-quantitative questionnaire has brought to light, for the first time, a more complete view of MF patients and their caregivers' experience, basing from their direct testimonies. Patients and caregivers' illness narratives, indeed, represent the more accurate way to follow their journey into the "falling ill phase", from the breakdown caused by the new health status, moving into the inner and more intimate world of the meaning of the disease. The good patients and caregivers' engagement in this project, independently from their age and educational qualification, allowed the identification of specific patterns of recurring thoughts, emotions and events. Through narratives, physicians have new elements to improve their understanding of patients and set up a more holistic approach. This is particularly relevant in the setting of chronic neoplasms, like myelofibrosis, that is projected to a relatively long survival but may be burdened by invalidating symptoms and reduced quality of life. Different perceptions between patients and physicians of the burden of illness and a poor patient-physician communication may delay or prevent the achievement of treatment goals [36].

The first indication from the present survey is the importance of a correct communication between the hematologist and the patient. Overall, patients would welcome a more gradual communication of the diagnosis along the several consecutive visits, thus entering slowly in the pathway of care. Language should be as simple as possible, focusing on patient-specific major clinical problems and possible solutions; prognosis should be addressed when required, explaining the impact of treatments and the limits of current risk scores in long-term prognostication.

The second consideration is related to the impact of relationships of care on the patients and caregivers' illness experience. In particular, narratives evidenced the key-role of the relationship between the hematologist and both patient and caregiver in enabling people to cope with MF. The successful relationship were based not only on technical expertise, but also on emotional empathy and communication skills in accordance to patients' wills, needs and possibilities.

The third message of this analysis relates to the crucial role of caregivers in the pathway of care in MF patients. Their heavy daily tasks lead to a high level of stress that could decrease through a more consideration of their needs and points of view, both in the organization of the supportive services and in the relationship of care. Medical teams should never underestimate caregivers' stress and loneliness, as they are part of the care. Several researches, indeed, have recognized the contribution of caregivers' perceptions of their experience on the quality of care provided [37].

Finally, one of the objectives of this study was to evaluate the burden of illness of MF from the patients and caregivers' perspective in Italy, with particular focus on social, welfare and economic aspects involved in the management of the disease. The enlarged concept of Burden of Illness allowed to move from the concept of "disease" to a more integrated concept of "illness", which considers the real out of pocket and daily costs, not only directly linked to the pathology, in the ordinary life. Findings from several articles suggest that the productivity costs due to intangible burden of illness associated with cancer are substantial [38]. Feedback from participants confirmed that direct health costs for patients and families are mostly covered by the National Health Service. However, MF revealed itself as a burden for patients and their families, with high intangible and indirect costs due to a progressive impairment of quality of life. Currently, the retirement age in Italy is gradually extending over time, as demonstrated by the fact that in this cohort of patients the median age was 65 years and 51% of them was still employed at the time of diagnosis of MF. As a consequence, limitations to daily activities led to a relevant loss of income in patients who were still in a productive age. The economic burden of illness of MF was calculated as high as 12.466 € per year from the patients' perspective, due to loss of family income; if the informal caregiver would have been replaced by a professional career, the family income loss would have risen to 20.326 €. These data are in agreement with a Spanish study [39] quantifying the burden of indirect and non-medical costs of MF as a total annual cost of 15.142 €. Furthermore, this survey shows that when the patient copes, there is a positive impact not only on QoL but also under the economic aspect, since the positive attitude allows to understand and improve the adherence to therapies, and to better effort working and daily activities.

The understanding of the resources of MF patients and their families revealed to be an important element to improve the relationships of care, the organization of the pathway and also to guide the economic choices to support people, allowing them to maintain their productivity and social role. Health care professionals and health care managers can benefit from the new metric offered by the integration of Narrative Medicine approach and Burden Of Illness tools, which allowed to reach these important results.

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