Research paper

Awareness of acute myeloid leukaemia risk induced by diagnosis of a myelodysplastic syndrome

Youssoufa M. Ousseine [a], Phyllis N. Butow [b], Claire Julian-Reynier [a,c], Rebecca Dring [d], Patrick Festy [e], Pierre Fenaux [f,g], Norbert Vey [c,h], Julien Mancini [a,i,*]

[a] Aix-Marseille Université, Inserm, IRD, UMR912, SESSTIM, "Cancers, Biomedicine & Society" group, Marseille F-13273 France
[b] Psycho-Oncology Co-operative Research Group (PoCoG) and Centre for Medical Psychology and Evidence-based Decision-making (CeMPED), University of Sydney, NSW 2006, Australia
[c] Institut Paoli-Calmettes, Marseille F-13273 France
[d] Leukaemia Foundation of Australia, Melbourne, Australia
[e] Connaître et Combattre les Myélodysplasies, Paris, France
[f] AP-HP, Hôpital Saint-Louis, Service d'Hématologie Clinique, Groupe Francophone des Myélodysplasies (GFM), Paris, France
[g] Paris 7 University, Paris, France
[h] Aix-Marseille Université, Inserm, CNRS, UM105, CRCM, Marseille F-13273, France
[i] APHM, Timone Hospital, Public Health Department (BioSTIC), Marseille F-13385, France

ARTICLE INFO

ABSTRACT

Myelodysplastic syndromes (MDS) can evolve to acute myeloid leukaemia (AML) in approximately 30% of cases. Knowing their AML risk is important for patients because it might impact adherence to care and psychological health. The aim of this study was to evaluate the awareness of AML risk among MDS patients and to study the factors associated with this awareness.

A self-administered questionnaire was mailed to all members of French and Australian patients' national MDS associations. Data of 301 patients were analysed.

Patients were satisfied with the information they had received, but 33.2% did not know that they had an increased risk of developing AML. Younger age, higher-risk MDS treatment, preferences for health-related information and satisfaction with information provided about treatment were the factors independently associated with awareness of AML risk. Compared to unaware patients, patients knowing their risk were more likely to participate in a hypothetical clinical trial (83.0% vs 72.4%, p = 0.043).

More efforts are needed to provide more systematic information about AML risk to patients wishing to know it. More research is needed to study if increasing awareness can lead to more active engagement of MDS patients in their care and can increase the rate of clinical trial participation.

© 2016 Published by Elsevier Ltd.

1. Introduction

Myelodysplastic syndromes (MDS) are a heterogeneous group of hematopoietic stem cell malignancies, mainly occurring in elderly persons with a median age at diagnosis of 70 years. MDS are characterized by peripheral blood cytopenias and by progression to acute myeloid leukaemia (AML) [1] in one third of patients [2]. This risk is highly heterogeneous depending on a variety of parameters such as the number and depth of cytopenias, marrow blast percentage, cytogenetics and more recently somatic mutations [3]. Risk of progression can be estimated using the International Prognostic Scoring System (IPSS), and its revised version (IPSS-R). However, even very low risk MDS are concerning, as 13% of the deaths in this subgroup in the IPSS-R database were due to AML [3].

In recent years, wide spread availability and accessibility of medical information for the general population, together with a stronger recognition of the importance of meeting patients' information needs, have led to a progressive change in patient-physician interactions: the past hierarchical model has moved towards a new cooperative model where most patients are told about and understand their diseases and the therapeutic possibilities. However, the heterogeneous and varied systems of MDS stratification make comprehension of MDS difficult for patients [4,5]. Patients' age, often greater than 70 years, and the complexity of the disease can add to comprehension difficulties. In such a context, the transmission

of information by the healthcare team should be tailored to the patient's health literacy level and information needs, to avoid information overload [6].

Among the different categories of information, prognostic issues are considered very important by heterogeneous cancer patients [7,8]. Discussing life expectancy has also been shown to increase cancer patients' satisfaction with care, and to decrease depression levels [9]. The manner of delivering prognostic information is important, impacting on patients' satisfaction, depression [10] and anxiety [11] Psychological profiles of patients are also determinants of this wish to be informed [12].

We recently conducted a survey [13] to evaluate MDS patients' information needs and their interest in a question prompt list, a simple tool listing questions they may wish to ask their doctor, which could help them to gain greater control over the flow of information. MDS patients were also particularly interested in knowing more about their prognosis. A major information item in this context is the increased risk of developing AML. Adequate awareness by patients suffering MDS of the risks induced by their disease, especially in terms of AML transformation and related risk of death, is important because it might be a determinant of adherence to follow-up and treatment if any (a majority of patients receive no treatment or only supportive care to correct one or several cytopenias associated with the MDS) [14]. Knowledge of these risks may also increase patients' willingness to participate in clinical trials. This would be consistent with results showing that women with node-positive breast cancer, who were aware of their future risk of relapse and death were more likely to consider participating in a clinical trial [15].

The aim of this study was to evaluate awareness of the AML risk in MDS patients and to study the factors associated with this awareness.

2. Methods

2.1. Study design

A cross-sectional survey was conducted with a broader objective of developing a question prompt list for MDS patients and family caregivers [13]. A self-administered questionnaire was mailed to all members of the Leukaemia Foundation of Australia patients' association and of the French CCM (*Connaître et Combattre les Myélodysplasies*) patients' association. In this survey, some caregivers were surveyed but the analyses presented here focused only on the subsample of 301 patients (171 French and 130 Australian) who responded.

2.2. Measures

2.2.1. Sociodemographic and psychological characteristics

Collected variables included socio-demographics (age, gender, marital status, occupational status, educational level, country, and first language) and psychological characteristics (preferences for health-related information, i.e. monitoring-blunting coping style [16] and MDS-related anxiety [17,18]).

2.2.2. Medical characteristics and experiences of the care

Physician who diagnosed the MDS, interval from MDS diagnosis and most recent treatments were collected. As in a previous survey [19] we did not ask for risk category (IPSS) because patients are often unaware of their score. Four categories of treatment were considered: watchful waiting, lower-risk MDS treatment (transfusions and hematopoietic growth factors, lenalidomide), higher-risk MDS treatment (5-azacitidine and chemotherapy including cytarabine injections), and stem cell transplant. Communication about prognosis was measured using two questions asking for perceived precision of communication and the wish for more/less information. Satisfaction with the information provided at MDS disclosure and about treatment was also collected. A specific question also asked for interest in clinical trial participation (*How likely would you be to participate in a clinical trial for the treatment of your MDS if it was offered to you?*).

2.2.3. Awareness of the AML risk

We evaluated the risk of AML progression using an ad hoc question: *Compared to someone of the same age and sex, do you think your MDS gives you a risk of developing leukaemia? 'More risk'* was considered the correct answer (i.e. adequate awareness) as even very low risk MDS might develop AML [3]. Other responses ('*Less risk*', '*Similar risk*', '*I don't know*') and non-responses were merged and considered as insufficient awareness.

2.2.4. Specific information needs

The specific information needs of patients were assessed by asking patients to rate (on a scale of 0–4) the usefulness of each of 53 questions in the question prompt list [13]. Ten dimensions emerged from factorial analysis: tests, prognosis, life expectancy, initial treatment, specific treatments, complementary treatments, side-effects, practical aspects and social issues.

2.3. Statistical analyses

Mean ± standard deviation (SD) and/or median [Interquartile range, IQR or minimum-maximum] were computed for every quantitative variable. Categorical variables were expressed as proportions. Student t-tests, Mann-Whitney tests and ANOVAs were used to compare continuous variables while chi-squared tests were used to compare categorical data. A binary logistic regression model was used to identify factors independently associated with insufficient awareness of leukaemia risk. Sensitivity analyses were performed. First, we excluded the non-response modality of the dependent variable, logistical model (non-responses [7.6%] were considered as insufficient awareness in the main analysis), results were unchanged. Second, an additional systematic adjustment for the duration of the disease (variable *Year from MDS diagnosis*) did not change the results.

To identify specific information needs dimensions, we used the Principal Component Analysis with varimax rotation. This exploratory method serves to highlight statistically latent structure of response regarding the usefulness of different questions patients could ask their doctors.

All statistical analyses were two-tailed and considered statistically significant when p-value < 0.05. They were performed using SPSS PASW Statistics 18.0 (IBM Inc., New York, USA).

3. Results

Patients' characteristics, surveyed a median of three years [IQR: 2–6 years] after MDS diagnosis, are displayed in Table 1. Most patients (73.3%) had high preferences for health-related information, i.e. a monitoring coping style. Patients were globally satisfied with the information they had received (Table 1), and satisfaction increased with disclosure of prognostic information (mean satisfaction ± SD: no talk about prognosis = 2.7 ± 1.0, vague talk about prognosis = 3.2 ± 0.9 and precise talk about prognosis = 3.7 ± 0.7, p < 0.001). However 33.2% (95% confidence interval 27.9% to 38.9%) of patients did not know that they had an increased risk of developing AML (1.7% thought they had less risk, 4.7% thought they had similar risk, 19.3% did not know, and 7.6% did not respond). Patients' awareness of this risk was the highest in patients who had a stem cell transplant (SCT) (Fig. 1); patients who had no treatment of their