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Research studies on patients' illness experience using the Narrative Medicine approach: a systematic review

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Complete List of Authors:	Fioretti, Chiara; Istituto Europeo di Oncologia, Applied Research Division for Cognitive and Psychological Science Mazzocco, Ketti; Istituto Europeo di Oncologia, Applied Research Division for Cognitive and Psychological Science; Università degli Studi di Milano, Department of Oncology and Hemato-oncology riva, silvia; Università degli Studi di Milano, Department of Oncology and Hemato-oncology Oliveri, Serena; Università degli Studi di Milano, Department of Oncology and Hemato-oncology Masiero, Marianna; Università degli Studi di Milano, Department of Oncology and Hemato-oncology Pravettoni, Gabriella; Istituto Europeo di Oncologia, Applied Research Division for Cognitive and Psychological Science; Università degli Studi di Milano, Department of Oncology and Hemato-oncology
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Research studies on patients’ illness experience using the Narrative Medicine approach: a systematic review

Fioretti Chiara¹, Mazzocco Ketti¹², Riva Silvia², Oliveri Serena², Masiero Marianna², Pravettoni Gabriella¹²

¹Applied Research Division for Cognitive and Psychological Science, European Institute of Oncology, Milan

²Department of Oncology and Emato-Oncology, University of Milan

Short Title: Narrative Medicine research studies: a systematic review

Corresponding Author:

Chiara Fioretti, Ph.D.
chiara.fioretti@ieo.it

Abstract

Objective: Since its birth about 30 years ago, the Narrative Medicine approach has increased in popularity in the medical context as well as in other disciplines. This paper aim to review Narrative Medicine research studies on patients' illness experience.

Setting and participants: MEDLINE, Psycinfo, EBSCO Psychological and Behavioural Science, The Chochrane Library and CINAHL databases were searched to identify all the research studies focused on the Narrative Medicine approach.

Primary and secondary outcome measures: number of participants, type of disease, race and age of participants, type of study, dependent variables, intervention methods, assessment.

Results: Of the 308 titles screened, we identified 10 research articles fitting inclusion criteria. Our systematic review showed that research on Narrative Medicine has no common specific methodology: narrative in Medicine is used as an intervention protocol as well as an assessment tool. Patients' characteristics, types of disease and data analysis procedures differ among the screened studies.

Conclusions: Narrative Medicine research in medical practice needs to find clear and specific protocols to deepen the impact of narrative on medical practice and on patients' lives.

Strengths and limitations: the manuscript proposes a first review of research studies in medicine conducted with a Narrative Medicine methodology. A systematic analysis was run to review scientific evidence in the field starting from the first publication on Narrative Medicine.

Due to the narrative nature of data, it was not possible to implement a met- analysis of the selected studies.

Keywords: Narrative Medicine; patient wellbeing; qualitative research; narrative intervention; systematic review

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3 1 **Introduction**
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Narrative Medicine (or Narrative Based Medicine) has developed as a theoretical and operative approach which has been increasingly discussed in recent years. This approach first came into existence about 30 years ago [1], and aims to introduce into daily medical practice the use of narrative as a tool to collect and interpret information on the patient’s experience of illness [2]. As Trisha Greenhalgh wrote (p. 318): “Clinical method is an interpretive act which draws on narrative skills to integrate the overlapping stories told by patients, clinicians, and test results”. Nevertheless, the current debate is focused more on the dualism between Narrative Medicine and Evidence-Based Medicine [3]: on the one hand medicine needs to be focused on scientifically-rigorous trials and to follow specific protocols, on the other the final aim of medical practice is always related to what a patient feels, what they perceive they feel and, above all, what they say they feel. What scholars have pointed out is that listening to the patient’s story is a tool to enrich not only the knowledge of their physical and psychological condition, but also to offer information with which to formulate the diagnosis [2; 4; 5; 6]. Thus, physicians and health staff need in their daily practice would seem to be to adopt a “Narrative Evidence Based Medicine” [7].

In this sense, numerous teaching programs in Narrative Medicine have recently been created (see <http://ce.columbia.edu/narrative-medicine> or <https://www.kcl.ac.uk/prospectus/graduate/medical-humanities>) to increase narrative competences of the health staff and to teach them how to use them in their daily work. Some studies have investigated the role of Narrative Medicine in teaching communication skills and increasing personal variables in health staff [8 ; 9].

Furthermore, experts in the field of Narrative Medicine argue that this approach also plays something of a therapeutic role for patients [10]. Thus, adopting a Narrative Medicine procedure in medical practice has positive consequences for the person who experiences a disease. In this case, the meaning of Narrative Medicine seems to be that of accompanying the patient through the listening to of her/his story of illness. Many authors such as Bury [11], have suggested that any

1 illness constitutes a disruption, a sort of discontinuance of an ongoing life. Lifespan psychologists
2 and developmental psychologists have stressed the importance of considering illness as a non-
3 normative transition of life which requests the individual to work towards the re-establishment of
4 the normative life balance. When a person faces a chronic illness, the need to reconstruct their life
5 story connecting the past life with the present experience of illness is strong. In this particular
6 context, narrative becomes an opportunity to give voice to the disruption and to provide it with a
7 time framework [12] not separated from the other life events which form part of the individual's
8 autobiographical story. There are many examples of the use of narrative to the repair life disruption
9 due to illness. Anatole Broyard, a renowned essayist of the New York Times, who in his
10 pathography reconstructed the story of his illness, famously affirmed that "storytelling seems to be
11 a natural reaction to illness. People bleed stories, and I've become a blood bank of them" [13].
12 In this domain, adopting a narrative-based approach could be a valid tool whereby patients are
13 helped to re-elaborate their experience. Nevertheless, scientific studies on the effectiveness of
14 Narrative Medicine and the evidence regarding its use in daily practice with patients have not been
15 reviewed so far.

16 This work aims to provide a systematic review of the research studies based on a Narrative
17 Medicine approach conducted with patients and/or with their caregivers. Thus, it aims to clarify the
18 scientific evidence in literature concerning the role of Narrative Medicine in patients' experience of
19 illness. Despite the importance that Narrative Medicine approach is acquiring in biomedical and
20 health sciences, just a few studies have tried to underline the scientific value of this approach.
21 Nevertheless, many studies have underlined the effect of narrative intervention in medicine. Zhou
22 and colleagues [14], for example, reviewed the randomized controlled trials assessing the effect of
23 Expressive Writing Intervention (EWI) [15] on health outcomes in breast cancer patients.
24 Comparing eleven articles, they found that EWI has a significant effect on reducing negative
25 somatic symptoms in a one to three-month follow-up.

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1 At the same time, Frisina and colleagues [16] explored the effects of the written Emotional
2 Disclosure paradigm on health outcomes of people with physical or psychiatric disorders. Meta-
3 analyzing nine articles, they found that expressive writing significantly improves health, but is more
4 effective on physical than on psychological health outcomes.
5 Studies on the effectiveness of narrative methods with patients have not focused solely on the
6 expressive writing paradigm. For instance, Cochinov and colleagues implemented a dignity therapy
7 intervention with patients near the end of life [17]. Asking patients to write a novel discussing
8 issues of their life story or those that they would most want remembered, they found in a test-retest
9 design a significant improvement of sense of dignity and a reduction of depression symptoms and
10 sense of suffering.
11 Furthermore, Houston and colleagues [18] assessed the use of storytelling as an intervention to
12 improve blood pressure. In a randomized trial study, they randomly divided patients suffering from
13 hypertension, using in the experimental group DVDs containing patient stories and measuring the
14 blood pressure 3, 6 and 9 months after the intervention. They found that the storytelling intervention
15 produced substantial and significant improvements in blood pressure for patients with baseline
16 uncontrolled hypertension [18].
17 Nevertheless, none of these studies considered their interventions based on narrative as part of the
18 Narrative Medicine approach. Although the procedures were based on the implementation of
19 narrative methods in medicine, authors did not refer to Narrative Medicine in their articles.
20 Since our aim was to review the role of studies on the Narrative Medicine approach itself, this
21 review took into consideration just those research studies that reported in the title or in the abstract
22 section the phrase Narrative Medicine (or Narrative Based Medicine). In extracting data and
23 organizing the review, we referred to the PRISMA Statement for reporting of systematic reviews
24 and meta-analyses [19].
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Materials and Methods

To find the most relevant articles for the systematic review, we searched main databases without restriction of language. We limited our search to begin from 1988, the year in which Kleinmann [1] published his first work on illness narratives. PubMed, PsycINFO, CINAHL, EBSCO Psychological and Behavioral Science and the Cochrane Library were employed. We did not handsearch conference proceeding or dissertations, due to limitations of time and resources. To select the most relevant works, we considered just the studies that met the following criteria: a) articles that reported the keywords “Narrative Medicine” or “Narrative Based Medicine” in the title or in the abstract; b) research studies, empirical or case study articles considered by authors as studies based on the Narrative Medicine approach. This second criterion was due to the need to separate Narrative Medicine studies from studies using narrative methods that were not considered by authors as works about Narrative Medicine [16; 14]. c) Research studies focused on patients or caregivers’ samples. After an initial literature review, the authors decided to include in the review process both intervention programs using a Narrative Medicine approach and research studies using it as a tool to explore patient’s experience of illness.

Two authors explored the scientific literature and independently extracted the data from every selected article. The general information (name of the authors and year of publication), as well as the study characteristics (type of study, dependent variables) and the participants characteristics (sample, disease, race, mean age of participants) were extracted (see Table 2). Since some of the selected articles reported interventions using a Narrative Medicine approach, we also extracted the intervention procedures as a variable of the review. When data were missing in the articles, we coded such data as “NR” (Not reported). See Table 1 for a summary of the search strategies and sources for the review implementation.

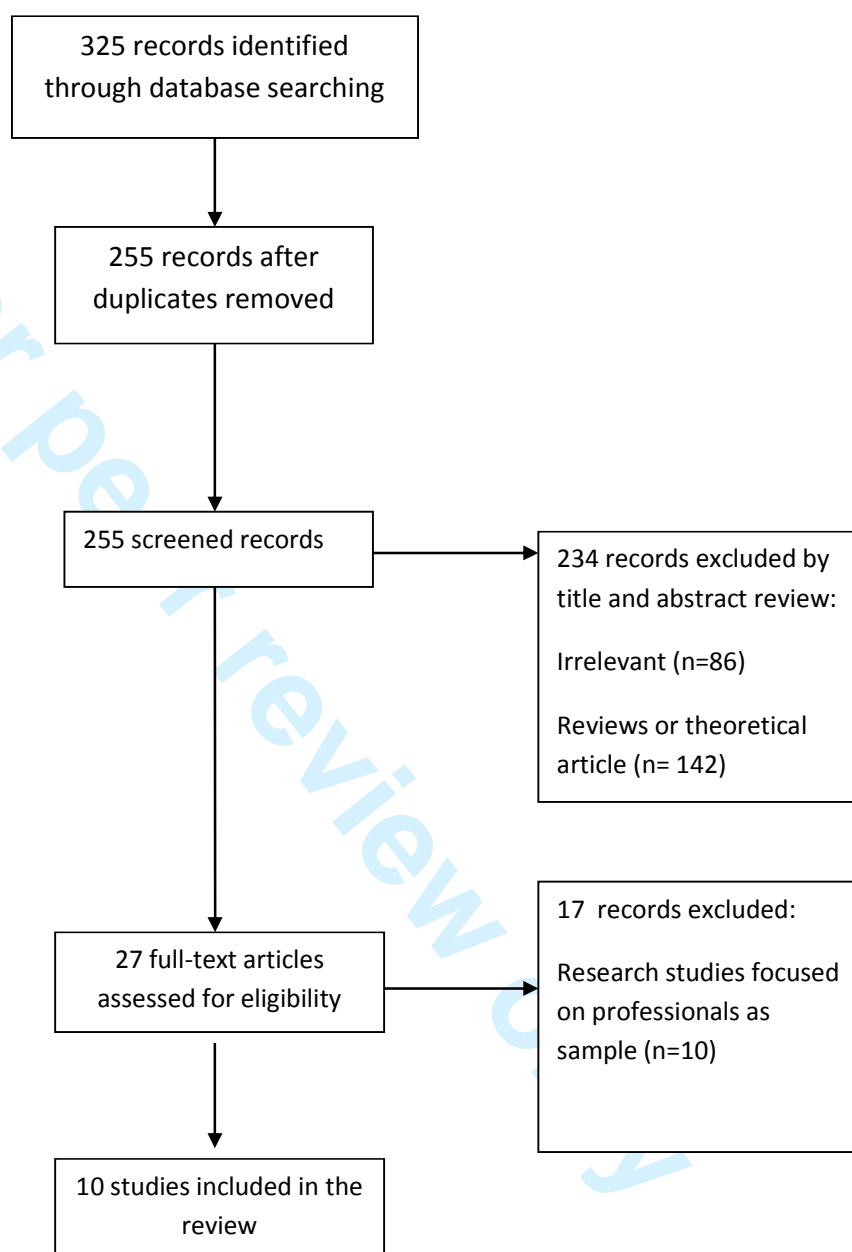
Table 1. Summary of search and source for the implementation review

Component	Description, inclusion/exclusion criteria and process of data extraction
Population	We included studies focused on patients' and/or caregivers' samples. Since scientific literature reported the use of a Narrative Medicine approach in every type of disease, we included in the review all the studies that considered patients affected by every kind of physical and mental illness. No restriction of race, age or other sample characteristics was considered.
Study design/type of study	Different types of study designs were considered, both research studies and intervention studies. Since studies adopting narrative methods are usually implemented with small size samples, we also included case studies in the review . We also selected studies considered by their own authors as part of the Narrative Medicine approach.
Dependent variables	Due to the characteristics of the checked articles, we included in the review research studies exploring different types of variables such as pain or well-being, satisfaction in participating in the intervention, and structure of illness narratives.
Databases	MEDLINE (186 records), CINAHL (69 records), EBSCO Psychological and Behavioural Science (30 records), The Cochrane Library (15 items) and PSYCINFO (25 items).
Other exclusion criteria	Dissertations, book reviews and editorials were not considered. Authors closely examined the bibliographies of the full-text screened articles to identify any additional possible studies

Results

Search results. The electronic literature search of articles was conducted in September 2015. Overall, a total of 325 abstracts and titles were analyzed, identifying 70 duplicates. Of the 255 titles and abstracts, 227 were excluded due to irrelevancy(n=85) or because they were reviews, theoretical or critical articles, editorials or book reviews (n= 142). The remaining 27 full-text articles were examined to identify the studies in line with all the inclusion criteria. 17 Articles were excluded if they described intervention projects or research studies conducted with professional staff. After having checked the reference lists of the selected full-texts, no additional items were found. Figure 1 illustrated the flow diagram of the present review.

Figure 1. Research studies on patients and caregivers conducted with the Narrative Medicine approach (2005/2015)



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1 *Included studies.* 10 studies considering the patients illness experience through a Narrative
2 Medicine approach were included in the systematic review. All the studies are presented and
3 described in Table 2.

For peer review only

1 **Table 2.** Characteristics of the included studies

Referen ce (year)	Sample	Disease	Race	Mean Age (SD)	Type of study	Dependent variables	Intervention methods	Assessme nt
Cepeda et al. (2008)	234 patients	Cancer	NR	48.5(12.4)	Randomiz ed trial	Pain and well-being perception	20 minutes narrative session one time per week for three weeks	Pain 0-10 scale and well- being Likert scale
Cotichell i (2012)	2 caregive rs	Pelizaeus- Merzbacher Disease (rare)	NR	NR	Case study	perception of socio- relational quality of service	NO	Analysis of categories emerged in narratives
Di Gangi et al. (2013)	332 caregive rs; 258 patients	NR (Intensive Care Unit)	NR	NR	Retrospect ive observatio nal	Patients' and caregivers' lives information derived from diary guestbooks	NO	cluster analysis with the software R Word Cloud 2.0 package
Donzelli et al. (2015)	3 patients	Pregnant after liver transplantation	NR	NR	Explorator y	Role of narrative medicine in facing illness experience	NO	narrative analysis of the collected text
Esquibel & Borkan (2014)	21 patients	NR	NR	NR	Explorator y	Chronic non cancer pain	NO	Thematic analysis of in- depth narrative interview
Greenhal gh et al. (2005)	NR	Diabetes	Asian	NR	Action- research	Learning on diabetes disease and change in behaviour	Storytelling groups with patients on their experiences of disease	Observati on of patients' behavior
Massimo & Zarri (2006)	50 patients	Pediatric leukemia or cancer		8(NR)		Children's perception of the disease	drawing therapy in hospital	Observati on of children's behavior
Michalak et al. (2014)	80 patients	Mental illness (bipolar disorder)	Cauca sian (83% of partici pants)	42.4(12.2)	Mixed methods design (prospecti ve, longitudin al)	Perception of Internalized stigma	Theatre class and performance. 6 weeks of 4h meetings, 3 to 4 times per week.	1. Day's Mental Illness Stigma Scale; 2. Internaliz ed Stigma of Mental Illness Scale; 4. Theatric performa nce evaluatio n

Smorti & Smorti (2013)	30 patients	Fertility problems/Assisted Reproduction Treatment	Caucasian	37(4.3)	Exploratory	Couple's psychological problems due to pregnancy via ART	NO	narrative analysis of common threads and phases of the parenthood transition
Wise et al. (2009)	11 patients	Cancer	Caucasian	67(NR)	Exploratory	Benefit from autobiographical storytelling	1.Telephone autobiographical interview 2. Manuscript on the patient life story 3. website to help people revise and share their story	In-depth interview focused on patients' experience with the intervention

Overall, a total amount of 1121 participants were involved in the considered studies. Of these, 761 were patients and 260 were caregivers. 155 patients participated in a randomized controlled trial as part of control group (n=78) or experimental group without Narrative Medicine intervention (n=77).

The majority of the participants were adults, while 50 of them were children.

Considering the type of disease experienced by the participants, three studies involved the participation of cancer patients; the others explored Pelizaeus-Merzbacher Disease, the experience of being pregnant after liver transplantation, diabetes, mental illness (bipolar disorder), fertility problems and Assisted Reproduction Treatment. Two studies did not report the participants' type of disease, but in one of these two authors reported that participants had been contacted in the intensive care unit of a hospital.

Out of ten studies, four described the individuals' race. In three studies, Caucasian patients participated in the research, in one case the study was conducted with the participation of Asian patients. The mean age was reported in five studies.

Considering the type of study conducted, results of systematic analysis underline different designs: four exploratory studies, one case study, one randomized trial, one action research, one retrospective observational study and one mixed methods design (prospective and longitudinal).

As mentioned above, the review includes both studies using Narrative Medicine as an intervention and studies using Narrative Medicine as a tool for collecting data.

Of the ten studies examined, five were intervention studies using Narrative Medicine procedures as an assessment tool. In those cases, the intervention procedures differed greatly among the studies.

Cepeda and colleagues [20] performed a randomized controlled trial in adult patients with cancer assessing whether using a Narrative Medicine approach decreases pain intensity and improves the global sense of well-being. They divided patients into three groups: the first (experimental group) wrote a story three times - once a week - for at least 20 minutes, about how cancer affected their lives; the second (attention group) completed a questionnaire on pain; the third (control group) attended weekly medical follow-up visits to receive the usual care provided for their therapies.

Overall, results showed that the three groups did not differ in pain intensity and well-being in the follow-up assessment. Nevertheless, patients who showed more emotional disclosure in narratives experienced significantly less pain and reported higher well-being scores than patients who wrote a narrative with a lower emotional disclosure.

Greenhalgh, Collard and Begum [21] used the Narrative Medicine approach as an intervention to promote learning about diabetes and behavioural change in Asian patients with diabetes. In an initial phase, researchers developed a storytelling training in a group of bilingual health advocates (BHAs). They then implemented a research activity in which trained BHAs set up storytelling groups for patients. Even though the study did not formally test the impact of the storytelling group on patients' blood glucose control or other psychological or medical variables, authors argued that after the intervention, patients reported being more confident and more active with respect to their illness.

Massimo and Zarri [22] performed an intervention on children suffering from cancer or leukemia focused on drawing therapy and aimed at reducing their stressful response to hospitalization and the dramatic changes in their lives. They collected both spontaneous and solicited drawings asking children "can you draw me a picture?" and later asking them to tell a brief story in their drawing. To assess patients' change, they observed drawings and children's behaviors and evaluated differences in the illness representation and in the subject of the drawings which emerged. Results underlined that the attention the children received made them more willing to cooperate, showing less stressful response to the hospitalization and disease therapies.

Michalak and colleagues [23] used theatre to address mental illness stigma in people with bipolar disorder. In a longitudinal study, they involved participants in theatre performance assessing stigma measures once before the intervention and twice after it (immediately and at follow-up three to four months later). The follow-up data collection also included an interview to elicit in-depth conversation of the participants' perceptions of the impact of the play on their mental stigma. The intervention consisted of six meetings of four hours each, conducted three to four times per week

1 and a final play with a 30-minutes question time. The intervention also involved the participations
2 of 84 healthcare providers. Since the aim of this review is to assess research studies and
3 intervention on patients and caregivers, we will not consider the results of this intervention
4 concerning the health staff. Overall, patients with bipolar disorders showed a small quantitative
5 change in mental stigma measures, with a significant decrease in the subscale of feelings of
6 alienation immediately after the performance but not in the follow-up. Conversely, comparing
7 quantitative data with the collected qualitative interviews, results showed that individuals expressed
8 continuous positive effects from the intervention.

9 Wise and colleagues [24] implemented an online narrative education program for 11 cancer patients
10 combining three types of intervention to help patients address emotional and existential issues. The
11 intervention was composed of: a) a telephone interview to elicit the life narrative; b) a life review
12 education with the final editing of a manuscript; c) a website giving instructional materials and
13 consultation to help people revising and sharing their story. The intervention effects were assessed
14 through in-depth exit interviews. Results showed that patients benefited from the intervention
15 appreciating the opportunity to capture their story and to engage families in its editing.

16 The other four studies included in the review used Narrative Medicine as part of the research
17 methodology implemented to evaluate different dependent variables.

18 Cotichelli [25] presented qualitative research published in an Italian journal using Narrative
19 Medicine to evaluate the perception of socio-relational quality of the health service in two parents
20 of a family facing a pediatric rare disease. Interviewing the two caregivers and implementing a
21 thematic analysis, the author found the following dimensions: a complex clinical context burdening
22 children and parents, the initial scarcity of helpful assistance and a close friendship network, the
23 limitations of the socio-sanitary services in diagnosing rare diseases and caring for children
24 suffering for those pathologies, the individual role of single professionals in providing support to
25 the families, the creation – in a following phase – of a support network, with a special role of the
26 voluntary associations.

Di Gangi and colleagues [26] explored the informative role of diaries and guest books in a narrative-based study. From 2009 and 2011 they collected stories spontaneously written by patients and caregivers attending the intensive care unit and implemented a software-based cluster analysis to identify the main themes. Results underlined that stories were frequently written in the form of a letter to patients to encourage them or to show emotional release. Diaries have been also used to provide feedback for the staff.

Donzelli and colleagues [27] explored the experience of pregnancy after liver transplantation using a Narrative Medicine tool approach. They conducted interviews and listened to the stories of three women who become pregnant after a liver transplantation, then they transcribed and analyzed the narrative plot to extrapolate the emerging themes. Three phases of the experience of illness were identified: a) the transplant, in which the mothers felt the need to talk about their operation; b) the pregnancy and the delivery, in which the mothers individuated the discovery of the pregnancy as the most delicate moment of their lives; finally the c) post-partum, in which the main protagonist of the story of disease is the child and a new prospect for the future.

Esquibel and Bokran [28] explored the ways in which chronic pain and opioid medication influence the doctor-patient relationship. To collect narratives they used a an in-depth interview with a semi-structured guide and open-ended questions. Researchers also interviewed patients' physicians, but results were not considered for the present review. The analysis of collected narratives revealed that patients focused their stories on suffering for chronic pain and on the role of opioid therapy to provide relief. The authors concluded arguing that the use of narrative to explore chronic pain has significant implications for improving the doctor-patient relationship.

Smorti and Smorti [29] used a Narrative-based Medicine approach to investigate medical success and couples' psychological problems in assisted reproduction treatment. They administered face-to-face semi-structured autobiographical interviews with couples to explore the story of the pregnancy in depth , transcribing the interviews verbatim and analyzing them via a thematic analysis. Results

showed that assisted reproduction treatment leads to a very stressful experience and is narrated by couples through a plot consisting of four phases: doubt, final sentence, victory and monitoring.

A final characteristic considered by the present review is the assessment procedure of the included studies. In this case, the review underlines that five studies assessed the dependent variables conducting narrative analysis of the collected texts. Among them, one study [26] implemented a text analysis software; the other authors carried out thematic, plot or narrative analysis.

Two studies assessed dependent variables using a Likert scale or self-report questionnaires: they conducted a quantitative and statistical analysis of collected data.

Two studies used observation of patients' behavior to assess changes in perception of disease and learning about disease management. Finally, Wise and colleagues [24] assessed the benefit from an autobiographical storytelling intervention through an in-depth interview .

Discussion

The main aim of the present work was that of reviewing the research/intervention studies adopting a Narrative Medicine approach with patients and caregivers. Overall, ten studies were included in the review. The main results to emerge provide evidence that Narrative Medicine is a useful tool to assess the patients' experience of illness and could be implemented in daily medical practice to enrich general clinical information focused on the needs and the critical aspects of patients' lives. This in turn could affect the normal therapeutic pathway.

Furthermore, Narrative Medicine also seems to be a powerful instrument for decreasing pain and increasing well-being related to illness (when patients' narratives show high emotional disclosure), for being more confident, active and cooperative in respect to the illness, for having a less stressful response and decreasing feelings of alienation, and finally for sharing illness stories with family members.

Although the debate within the Narrative Medicine approach started some thirty years ago, the systematic review shows that the majority of scientific literature in the field is still composed of

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1 theoretical articles or critical reviews. Furthermore, all the studies included in the review were
2 conducted in the last 10 years. These data stress the need for implementing more studies on the
3 effects and the power of Narrative Medicine on patients' experience of illness.
4 Particularly, data collection underlines a continuity among studies on Narrative Medicine and
5 studies on other theoretical approaches such as Expressive Writing paradigm, Emotional Disclosure
6 and Dignity Therapy in palliative care. All these studies use the storytelling as a tool to help patients
7 to express their feelings, worries and doubts about the disease. It is interesting to note, for instance,
8 the similarity between the study of Michalak and colleagues [23] on the use of theatre to address the
9 stigma of mental illness and the study of Roberts and colleagues [30] on an intervention program in
10 adolescents and young adults based on applied drama and theatrical performance. Both studies
11 demonstrated the role of theatre in decreasing mental stigma, even though the first focused on
12 bipolar disorder adult patients [23] and the second on early psychosis in adolescence and emerging
13 adulthood [30]. Nevertheless, Roberts and colleague did not recognize their work as a Narrative
14 Medicine study.
15 As we mentioned earlier, there are also many similarities among Narrative Medicine and emotional
16 disclosure and expressive writing interventions. The study of Cepeda and colleagues [20], for
17 instance, adopted an intervention focused on emotional disclosure through writing once a week for
18 three weeks, about doubts, fears, feelings related to the disease. This intervention seems to be very
19 similar to studies reviewed by Smith [31] on written emotional expression, although the latter did
20 not refer in his review to a Narrative Medicine approach.
21 In this sense, it seems very opportune to define the boundaries of the Narrative Medicine approach
22 in order to give it a scientific independence and common protocols to implement.
23 Another important topic that emerged from the review is that studies on Narrative Medicine have
24 used this approach both as an intervention and as a tool to collect narrative data. From our point of
25 view, these two different types of Narrative Medicine study also imply different research goals.

1 The intervention studies included in the present review were focused on the use of Narrative
2 Medicine to collect information on the effectiveness of this approach and on the patient benefits
3 which derived from it. In this sense, the main aim of the intervention studies on Narrative Medicine
4 seems to be that of assessing the efficacy of the use of the approach on patients: data collected
5 would be evidence to spur health staff into using it in their daily practice.

6 If instead we look at studies which underline Narrative Medicine as an instrument for collecting
7 narrative data we see that these aim to stress the importance of the approach for providing
8 qualitative information on the patient's experience of their illness experience to the health staff.

9 Thus, data collected would provide practical knowledge to take into account in medical practice.

10 For instance, physicians caring for women who have undergone transplantation should take into
11 consideration that the discovery of the pregnancy is a very delicate moment in their patients' lives
12 and calls for particular attention to, and organization of, healthcare [27]. Greenhalgh and Hurwitz
13 [5], in fact, argued that "narratives offer a method for addressing existential qualities such as inner
14 hurt, despair, hope, grief, and moral pain which frequently accompany, and may even constitute,
15 people's illnesses" (p. 318).

16 Considering the procedural limitations of reviewed works, the systematic review has shown that
17 studies have engaged the participation of very different samples for size, patients' main age and
18 type of disease. Considering that every disease has its own individual care path and thus is
19 associated with different physical and psychological experiences, depending on patients'
20 characteristics, it seems opportune to take into account the individual disease in the design planning
21 of Narrative Medicine studies with patients. More information on the type of disease and the sample
22 characteristics should be addressed in future studies.

23 Furthermore, intervention studies overall did not report the integration of control groups in their
24 research design. Except for the randomized trial conducted by Cepeda and colleagues [20], all other
25 studies have investigated the role of Narrative Medicine interventions without comparing them with
26 other interventions or control groups. Also, the nature of the intervention greatly varies. Participants

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1 in the studies included in the review have performed telephone autobiographical interviews,
2 manuscript editing, theatre performance, drawing, storytelling groups, and twenty-minute narrative
3 sections.
4 Another limitation is the high variety of data coding of narratives collected in the research studies
5 using Narrative Medicine as a tool for collecting patients’ experiences. In one case [26] researches
6 implemented a software of textual analysis assessing the emerged clusters of narratives, in the other
7 studies authors preferred to conduct a thematic, plot, or narrative analysis.
8 To conclude, it seems very suitable to define the boundaries of the Narrative Medicine approach
9 when it is used in research with patients in order to give it scientific independence and common
10 protocols to implement. Thus, intervention programs should be compliant with the theoretical
11 framework, as well as the analysis of patients’ experiences collecting through a Narrative Medicine
12 approach. Starting from the copious scientific literature on the topic, researchers should find a
13 common methodology and a shared procedure which will give the opportunity to replicate the study
14 in other contexts and with patients suffering from different diseases [32].

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Contributors

The review was conceived by CF and KM. Data extraction was carried out by SR, MM and SO with support from CF and KM. Reporting of findings was led by CF with support from KM and GP. All authors contributed to manuscript preparation and approved the final version.

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Declaration of competing interests

We have read and understood BMJ policy on declaration of interests and declare that we have no competing interests

Data sharing statement

No additional data available

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PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	3-5
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	7
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	6
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	6
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	6
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	6
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2 for each meta-analysis).	

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PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	7
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	9-16
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	9-16
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	9-16
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	16-17
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	18-19
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	18-19
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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Keywords:	Narrative Medicine, patient wellbeing, QUALITATIVE RESEARCH, narrative intervention, systematic review

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Research studies on patients’ illness experience using the Narrative Medicine approach: a systematic review

Fioretti Chiara¹, Mazzocco Ketti¹², Riva Silvia², Oliveri Serena², Masiero Marianna², Pravettoni Gabriella¹²

¹Applied Research Division for Cognitive and Psychological Science, European Institute of Oncology, Milan

²Department of Oncology and Hemato-Oncology, University of Milan

Short Title: Narrative Medicine research studies: a systematic review

Corresponding Author:

Chiara Fioretti, Ph.D.
chiara.fioretti@ieo.it

Abstract

Objective: Since its birth about 30 years ago, the Narrative Medicine approach has increased in popularity in the medical context as well as in other disciplines. This paper aims to review Narrative Medicine research studies on patients' and their caregivers' illness experience.

Setting and participants: MEDLINE, Psycinfo, EBSCO Psychological and Behavioural Science, The Chochrane Library and CINAHL databases were searched to identify all the research studies focused on the Narrative Medicine approach reporting in the title, in the abstract and in the keywords the words "Narrative Medicine" or "Narrative based Medicine".

Primary and secondary outcome measures: number of participants, type of disease, race and age of participants, type of study, dependent variables, intervention methods, assessment.

Results: Of the 325 titles screened, we identified 10 research articles fitting inclusion criteria. Our systematic review showed that research on Narrative Medicine has no common specific methodology: narrative in Medicine is used as an intervention protocol as well as an assessment tool. Patients' characteristics, types of disease and data analysis procedures differ among the screened studies.

Conclusions: Narrative Medicine research in medical practice needs to find clear and specific protocols to deepen the impact of narrative on medical practice and on patients' lives.

Strengths and limitations: the manuscript proposes a first review of research studies in medicine conducted with a Narrative Medicine methodology. A systematic analysis was run to review scientific evidence in the field starting from the first publication on Narrative Medicine.

Due to the narrative nature of data, it was not possible to implement a meta-analysis of the selected studies.

Keywords: Narrative Medicine; patient wellbeing; qualitative research; narrative intervention; systematic review

Introduction

Narrative Medicine (or Narrative Based Medicine) has developed as a theoretical and operative approach which has been increasingly discussed in recent years. This approach first came into existence about 30 years ago [1], and aims to introduce into daily medical practice the use of narrative as a tool to collect and interpret information on the patient’s experience of illness [2]. As Trisha Greenhalgh wrote (p. 318): “Clinical method is an interpretive act which draws on narrative skills to integrate the overlapping stories told by patients, clinicians, and test results”. Nevertheless, the current debate is focused more on the dualism between Narrative Medicine and Evidence-Based Medicine [3]: on the one hand medicine needs to be focused on scientifically-rigorous trials and to follow specific protocols, on the other the final aim of medical practice is always related to what a patient feels, what they perceive they feel and, above all, what they say they feel. What scholars have pointed out is that listening to the patient’s story is a tool to enrich not only the knowledge of their physical and psychological condition, but also to offer information with which to formulate the diagnosis [2; 4; 5; 6]. Thus, physicians and health staff need in their daily practice would seem to be to adopt a “Narrative Evidence Based Medicine” [7].

In this sense, numerous teaching programs in Narrative Medicine have recently been created (see <http://ce.columbia.edu/narrative-medicine> or <https://www.kcl.ac.uk/prospectus/graduate/medical-humanities>) to increase narrative competences of the health staff and to teach them how to use them in their daily work. Some studies have investigated the role of Narrative Medicine in teaching communication skills and increasing personal variables in health staff [8 ; 9].

Furthermore, experts in the field of Narrative Medicine argue that this approach also plays something of a therapeutic role for patients [10]. Thus, adopting a Narrative Medicine procedure in medical practice has positive consequences for the person who experiences a disease. In this case, the meaning of Narrative Medicine seems to be that of accompanying the patient through the listening of her/his story of illness. Many authors such as Bury [11], have suggested that any illness

1 constitutes a disruption, a sort of discontinuance of an ongoing life. Lifespan psychologists and
2 developmental psychologists have stressed the importance of considering illness as a non-normative
3 transition of life which requests the individual to work towards the re-establishment of the
4 normative life balance. When a person faces a chronic illness, the need to reconstruct their life story
5 connecting the past life with the present experience of illness is strong. In this particular context,
6 narrative becomes an opportunity to give voice to the disruption and to provide it with a time
7 framework [12] not separated from the other life events which form part of the individual's
8 autobiographical story. There are many examples of the use of narrative to the repair life disruption
9 due to illness. Anatole Broyard, a renowned essayist of the New York Times, who in his
10 pathography reconstructed the story of his illness, famously affirmed that "storytelling seems to be
11 a natural reaction to illness. People bleed stories, and I've become a blood bank of them" [13].
12 In this domain, adopting a narrative-based approach could be a valid tool whereby patients are
13 helped to re-elaborate their experience. Nevertheless, scientific studies on the effectiveness of
14 Narrative Medicine and the evidence regarding its use in daily practice with patients have not been
15 reviewed so far.

16 This work aims to provide a systematic review of the research studies based on a Narrative
17 Medicine approach conducted with patients and/or with their caregivers. Thus, it aims to clarify the
18 scientific evidence in literature concerning the role of Narrative Medicine in patients' experience of
19 illness. Despite the importance that Narrative Medicine approach is acquiring in biomedical and
20 health sciences, just a few studies have tried to underline the scientific value of this approach.
21 Nevertheless, many studies have underlined the effect of narrative intervention in medicine. Zhou
22 and colleagues [14], for example, reviewed the randomized controlled trials assessing the effect of
23 Expressive Writing Intervention (EWI) [15] on health outcomes in breast cancer patients. This kind
24 of intervention, asking participants to reflect in a written format about negative past life events,
25 aims to improve emotional expression and elaboration of stressful situations improving

psychological and physical health. Comparing eleven articles, they found that EWI has a significant effect on reducing negative somatic symptoms in a one to three-month follow-up. At the same time, Frisina and colleagues [16] explored the effects of the written Emotional Disclosure paradigm on health outcomes of people with physical or psychiatric disorders. Meta-analyzing nine articles, they found that expressive writing significantly improves health, but is more effective on physical than on psychological health outcomes. Studies on the effectiveness of narrative methods with patients have not focused solely on the expressive writing paradigm. For instance, Cochinov and colleagues implemented a dignity therapy intervention with patients near the end of life [17]. Asking patients to write a novel discussing issues of their life story or those that they would most want remembered, they found in a test-retest design a significant improvement of sense of dignity and a reduction of depression symptoms and sense of suffering. Furthermore, Houston and colleagues [18] assessed the use of storytelling as an intervention to improve blood pressure. In a randomized trial study, they randomly divided patients suffering from hypertension, using in the experimental group DVDs containing patient stories and measuring the blood pressure 3, 6 and 9 months after the intervention. They found that the storytelling intervention produced substantial and significant improvements in blood pressure for patients with baseline uncontrolled hypertension [18]. Nevertheless, none of these studies considered their interventions based on narrative as part of the Narrative Medicine approach. Although the procedures were based on the implementation of narrative methods in medicine, authors did not refer to Narrative Medicine in their articles. Since our aim was to review the role of studies on the Narrative Medicine approach itself, this review took into consideration just those research studies that reported in the title, in the abstract section or in the keywords the phrase Narrative Medicine (or Narrative Based Medicine). In extracting data and organizing the review, we referred to the PRISMA Statement for reporting of systematic reviews and meta-analyses [19].

Materials and Methods

To find the most relevant articles for the systematic review, we searched main databases without restriction of language. We limited our search to begin from 1988, the year in which Kleinmann [1] published his first work on illness narratives. PubMed, PsycINFO, CINAHL, EBSCO Psychological and Behavioral Science and the Cochrane Library were employed. We did not handsearch conference proceeding or dissertations, due to limitations of time and resources. To select the most relevant works, we considered just the studies that met at least one of the following criteria: a) articles that reported the words “Narrative Medicine” or “Narrative Based Medicine” in the title, in the abstract and/or in the keywords; b) research studies, empirical or case study articles referring to the Narrative Medicine approach in the background and/or methodological section. This second criterion was due to the need to separate Narrative Medicine studies from studies using narrative methods that were not considered by authors as works about Narrative Medicine [16; 14]. c) Research studies focused on patients or caregivers’ samples. After an initial literature review, the authors decided to include in the review process both intervention programs using a Narrative Medicine approach and research studies using it as a tool to explore patient’s experience of illness. Two authors explored the scientific literature and independently extracted the data from every selected article. See Table 1 for a summary of the search strategies and sources for the review implementation. As an example, for the PubMed database the used search string was: ((Narrative Medicine[Title/Abstract]) OR Narrative based Medicine[Title/Abstract]) AND ("1988"[Date - Publication] : "3000"[Date - Publication]).

The general information (name of the authors and year of publication), as well as the study characteristics (type of study, dependent variables) and the participants characteristics (sample, disease, race, mean age of participants) were extracted (see Table 2). Since some of the selected articles reported interventions using a Narrative Medicine approach, we also extracted the intervention procedures as a variable of the review. When data were missing in the articles, we coded such data as “NR” (Not reported). Since Narrative Medicine is a theoretical but also

methodological approach providing clinicians and researchers with intervention tool in their daily activity with patients, authors assessed and graded the quality of included studies focusing on their methodological design and on the dependent variables investigated.

Table 1. Summary of search and source for the implementation review

Component	Description, inclusion/exclusion criteria and process of data extraction
Population	We included studies focused on patients' and/or caregivers' samples. Since scientific literature reported the use of a Narrative Medicine approach in every type of disease, we included in the review all the studies that considered patients affected by every kind of physical and mental illness. No restriction of race, age or other sample characteristics was considered.
Study design/type of study	Different types of study designs were considered, both research studies and intervention studies. Since studies adopting narrative methods are usually implemented with small size samples, we also included case studies in the review. We also selected studies considered by their own authors as part of the Narrative Medicine approach.
Dependent variables	Due to the characteristics of the checked articles, we included in the review research studies exploring different types of variables such as pain or well-being, satisfaction in participating in the intervention, and structure of illness narratives.
Databases	PubMed (186 records), CINAHL (69 records), EBSCO Psychological and Behavioural Science (30 records), The Cochrane Library (15 items) and PSYCINFO (25 items). Authors closely examined the bibliographies of the full-text screened articles to identify any additional possible study
Other exclusion criteria	Dissertations, book reviews and editorials were not considered.

Results

Search results. The electronic literature search of articles was conducted in September 2015. Overall, a total of 325 abstracts and titles were analyzed, identifying 70 duplicates. Of the 255 titles and abstracts, 228 were excluded due to irrelevancy (n=86) or because they were reviews, theoretical or critical articles, editorials or book reviews (n= 142). The remaining 27 full-text articles were examined to identify the studies in line with all the inclusion criteria. 17 Articles were excluded if they described intervention projects or research studies conducted with professional

1 staff. After having checked the reference lists of the selected full-texts, no additional items were
2 found. Figure 1 illustrated the flow diagram of the present review.

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4 *Included studies.* 10 studies considering the patients illness experience through a Narrative
5 Medicine approach were included in the systematic review. All the studies are presented and
6 described in Table number 2.

Table 2. Characteristics of the included studies

Referen ce (year)	Sample	Disease	Race	Mean Age (SD)	Type of study	Dependent variables	Intervention methods	Assessme nt
Cepeda et al. (2008)	234 patients	Cancer	NR	48.5(12.4)	Randomiz ed trial	Pain and well-being perception	20 minutes narrative session one time per week for three weeks	Pain 0-10 scale and well- being Likert scale
Cotichell i (2012)	2 caregive rs	Pelizaeus- Merzbacher Disease (rare)	NR	NR	Case study	perception of socio- relational quality of service	NO	Analysis of categories emerged in narratives
Di Gangi et al. (2013)	332 caregive rs; 258 patients	NR (Intensive Care Unit)	NR	NR	Retrospect ive observatio nal	Patients' and caregivers' lives information derived from diary guestbooks	NO	cluster analysis with the software R Word Cloud 2.0 package
Donzelli et al. (2015)	3 patients	Pregnant after liver transplantation	NR	NR	Explorator y	Role of narrative medicine in facing illness experience	NO	narrative analysis of the collected text
Esquibel & Borkan (2014)	21 patients	NR	NR	NR	Explorator y	Chronic non cancer pain	NO	Thematic analysis of in- depth narrative interview
Greenhal gh et al. (2005)	NR	Diabetes	Asian	NR	Action- research	Learning on diabetes disease and change in behaviour	Storytelling groups with patients on their experiences of disease	Observati on of patients' behavior
Massimo & Zarri (2006)	50 patients	Pediatric leukemia or cancer		8(NR)		Children's perception of the disease	drawing therapy in hospital	Observati on of children's behavior
Michalak et al. (2014)	80 patients	Mental illness (bipolar disorder)	Cauca sian (83% of partici pants)	42.4(12.2)	Mixed methods design (prospecti ve, longitudin al)	Perception of Internalized stigma	Theatre class and performance. 6 weeks of 4h meetings, 3 to 4 times per week.	1. Day's Mental Illness Stigma Scale; 2. Internaliz ed Stigma of Mental Illness Scale; 4. Theatric performa nce evaluatio n

Smorti & Smorti (2013)	30 patients	Fertility problems/Assisted Reproduction Treatment	Caucasian	37(4.3)	Exploratory	Couple's psychological problems due to pregnancy via ART	NO	narrative analysis of common threads and phases of the parenthood transition
Wise et al. (2009)	11 patients	Cancer	Caucasian	67(NR)	Exploratory	Benefit from autobiographical storytelling	1. Telephone autobiographical interview 2. Manuscript on the patient life story 3. website to help people revise and share their story	In-depth interview focused on patients' experience with the intervention

Overall, a total amount of 1021 participants were involved in the considered studies. Of these, 687 were patients and 334 were caregivers. 155 patients participated in a randomized controlled trial as part of control group (n=78) or experimental group without Narrative Medicine intervention (n=77). The majority of the participants were adults, while 50 of them were children. Considering the type of disease experienced by the participants, three studies involved the participation of cancer patients; the others explored Pelizaeus-Merzbacher Disease, the experience of being pregnant after liver transplantation, diabetes, mental illness (bipolar disorder), fertility problems and Assisted Reproduction Treatment. Two studies did not report the participants' type of disease, but in one of these two, authors reported that participants had been contacted in the intensive care unit of a hospital. Out of ten studies, four described the individuals' race. In three studies, Caucasian patients participated in the research, in one case the study was conducted with the participation of Asian patients. The mean age was reported in five studies.

1 Considering the type of study conducted, results of systematic analysis underline different designs:
2 four exploratory studies, one case study, one randomized trial, one action research, one
3 retrospective observational study and one mixed methods design (prospective and longitudinal).
4 As mentioned above, the review includes both studies using Narrative Medicine as an intervention
5 and studies using Narrative Medicine as a tool for collecting data.
6 Of the ten studies examined, five were intervention studies using Narrative Medicine procedures as
7 an assessment tool. In those cases, the intervention procedures differed greatly among the studies.
8 Cepeda and colleagues [20] performed a randomized controlled trial in adult patients with cancer
9 assessing whether using a Narrative Medicine approach decreases pain intensity and improves the
10 global sense of well-being. They divided patients into three groups: the first (experimental group)
11 wrote a story three times - once a week - for at least 20 minutes, about how cancer affected their
12 lives; the second (attention group) completed a questionnaire on pain; the third (control group)
13 attended weekly medical follow-up visits to receive the usual care provided for their therapies.
14 Overall, results showed that the three groups did not differ in pain intensity and well-being in the
15 follow-up assessment. Nevertheless, patients who showed more emotional disclosure in narratives
16 experienced significantly less pain and reported higher well-being scores than patients who wrote a
17 narrative with a lower emotional disclosure.
18 Greenhalgh, Collard and Begum [21] used the Narrative Medicine approach as an intervention to
19 promote learning about diabetes and behavioural change in Asian patients with diabetes. In an
20 initial phase, researchers developed a storytelling training in a group of bilingual health advocates
21 (BHAs). They then implemented a research activity in which trained BHAs set up storytelling
22 groups for patients. Even though the study did not formally test the impact of the storytelling group
23 on patients' blood glucose control or other psychological or medical variables, authors argued that
24 after the intervention, patients reported being more confident and more active with respect to their
25 illness.

Massimo and Zarri [22] performed an intervention on children suffering from cancer or leukemia focused on drawing therapy and aimed at reducing their stressful response to hospitalization and the dramatic changes in their lives. They collected both spontaneous and solicited drawings asking children “can you draw me a picture?” and later asking them to tell a brief story in their drawing. To assess patients’ change, they observed drawings and children’s behaviors and evaluated differences in the illness representation and in the subject of the drawings which emerged. Results underlined that the attention the children received made them more willing to cooperate, showing less stressful response to the hospitalization and disease therapies.

Michalak and colleagues [23] used theatre to address mental illness stigma in people with bipolar disorder. In a longitudinal study, they involved participants in theatre performance assessing stigma measures once before the intervention and twice after it (immediately and at follow-up three to four months later). The follow-up data collection also included an interview to elicit in-depth conversation of the participants’ perceptions of the impact of the play on their mental stigma. The intervention consisted of six meetings of four hours each, conducted three to four times per week and a final play with a 30-minutes question time. The intervention also involved the participations of 84 healthcare providers. Since the aim of this review is to assess research studies and intervention on patients and caregivers, we will not consider the results of this intervention concerning the health staff. Overall, patients with bipolar disorders showed a small quantitative change in mental stigma measures, with a significant decrease in the subscale of feelings of alienation immediately after the performance but not in the follow-up. Conversely, comparing quantitative data with the collected qualitative interviews, results showed that individuals expressed continuous positive effects from the intervention.

Wise and colleagues [24] implemented an online narrative education program for 11 cancer patients combining three types of intervention to help patients address emotional and existential issues. The intervention was composed of: a) a telephone interview to elicit the life narrative; b) a life review education with the final editing of a manuscript; c) a website giving instructional materials and

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3 1 consultation to help people revising and sharing their story. The intervention effects were assessed
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5 2 through in-depth exit interviews. Results showed that patients benefited from the intervention
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7 3 appreciating the opportunity to capture their story and to engage families in its editing.
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10 4 The other four studies included in the review used Narrative Medicine as part of the research
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12 5 methodology implemented to evaluate different dependent variables.
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14 6 Cotichelli [25] presented qualitative research published in an Italian journal using Narrative
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16 7 Medicine to evaluate the perception of socio-relational quality of the health service in two parents
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18 8 of a family facing a pediatric rare disease. Interviewing the two caregivers and implementing a
19
20 9 thematic analysis, the author found the following dimensions: a complex clinical context burdening
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22 10 children and parents, the initial scarcity of helpful assistance and a close friendship network, the
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24 11 limitations of the socio-sanitary services in diagnosing rare diseases and caring for children
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26 12 suffering for those pathologies, the individual role of single professionals in providing support to
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28 13 the families, the creation – in a following phase – of a support network, with a special role of the
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30 14 voluntary associations.
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33 15 Di Gangi and colleagues [26] explored the informative role of diaries and guest books
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35 16 in a narrative-based study. From 2009 and 2011 they collected stories spontaneously written by
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37 17 patients and caregivers attending the intensive care unit and implemented a software-based cluster
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39 18 analysis to identify the main themes. Results underlined that stories were frequently written in the
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41 19 form of a letter to patients to encourage them or to show emotional release. Diaries have been also
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43 20 used to provide feedback for the staff.
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46 21 Donzelli and colleagues [27] explored the experience of pregnancy after liver transplantation using
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48 22 a Narrative Medicine tool approach. They conducted interviews and listened to the stories of three
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50 23 women who become pregnant after a liver transplantation, then they transcribed and analyzed the
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52 24 narrative plot to extrapolate the emerging themes. Three phases of the experience of illness were
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54 25 identified: a) the transplant, in which the mothers felt the need to talk about their operation; b) the
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56 26 pregnancy and the delivery, in which the mothers individuated the discovery of the pregnancy as the
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1 most delicate moment of their lives; finally the c) post-partum, in which the main protagonist of the
2 story of disease is the child and a new prospect for the future.

3 Esquibel and Bokran [28] explored the ways in which chronic pain and opioid medication influence
4 the doctor-patient relationship. To collect narratives they used an in-depth interview with a semi-
5 structured guide and open-ended questions. Researchers also interviewed patients' physicians, but
6 results were not considered for the present review. The analysis of collected narratives revealed that
7 patients focused their stories on suffering for chronic pain and on the role of opioid therapy to
8 provide relief. The authors concluded arguing that the use of narrative to explore chronic pain has
9 significant implications for improving the doctor-patient relationship.

10 Smorti and Smorti [29] used a Narrative-based Medicine approach to investigate medical success
11 and couples' psychological problems in assisted reproduction treatment. They administered face-to-
12 face semi-structured autobiographical interviews with couples to explore the story of the pregnancy
13 in depth, transcribing the interviews verbatim and analyzing them via a thematic analysis. Results
14 showed that assisted reproduction treatment leads to a very stressful experience and is narrated by
15 couples through a plot consisting of four phases: doubt, final sentence, victory and monitoring.

16 A final characteristic considered by the present review is the assessment procedure of the included
17 studies. In this case, the review underlines that five studies assessed the dependent variables
18 conducting narrative analysis of the collected texts. Among them, one study [26] implemented a
19 text analysis software; the other authors carried out thematic, plot or narrative analysis.

20 Two studies assessed dependent variables using a Likert scale or self-report questionnaires: they
21 conducted a quantitative and statistical analysis of collected data.

22 Two studies used observation of patients' behavior to assess changes in perception of disease and
23 learning about disease management. Finally, Wise and colleagues [24] assessed the benefit from an
24 autobiographical storytelling intervention through an in-depth interview.

Discussion

The main aim of the present work was that of reviewing the research/intervention studies adopting a Narrative Medicine approach with patients and caregivers. Overall, ten studies were included in the review. The main results emerged provide evidence that Narrative Medicine is a useful tool to assess the patients' experience of illness and could be implemented in daily medical practice to enrich general clinical information focused on the needs and the critical aspects of patients' lives. This in turn could affect the normal therapeutic pathway.

Furthermore, Narrative Medicine also seems to be a powerful instrument for decreasing pain and increasing well-being related to illness (when patients' narratives show high emotional disclosure), for being more confident, active and cooperative in respect to the illness, for having a less stressful response and decreasing feelings of alienation, and finally for sharing illness stories with family members.

Although the debate within the Narrative Medicine approach started some thirty years ago, the systematic review shows that the majority of scientific literature in the field is still composed of theoretical articles or critical reviews. Furthermore, all the studies included in the review were conducted in the last 10 years. These data stress the need for implementing more studies on the effects and the power of Narrative Medicine on patients' experience of illness.

Particularly, data collection underlines a continuity among studies on Narrative Medicine and studies on other theoretical approaches such as Expressive Writing/Emotional Disclosure paradigm and Dignity Therapy in palliative care. All these studies use the storytelling as a tool to help patients to express their feelings, worries and doubts about the disease. It is interesting to note, for instance, the similarity between the study of Michalak and colleagues [23] on the use of theatre to address the stigma of mental illness and the study of Roberts and colleagues [30] on an intervention program in adolescents and young adults based on applied drama and theatrical performance. Both studies demonstrated the role of theatre in decreasing mental stigma, even though the first focused on bipolar disorder adult patients [23] and the second on early psychosis in adolescence and emerging

adulthood [30]. Nevertheless, Roberts and colleague did not recognize their work as a Narrative Medicine study.

As we mentioned earlier, there are also many similarities among Narrative Medicine and emotional disclosure/expressive writing interventions. The study of Cepeda and colleagues [20], for instance, adopted an intervention focused on emotional disclosure through writing once a week for three weeks, about doubts, fears, feeling related to the disease. This intervention seems to be very similar to studies reviewed by Smith [31] on written emotional expression, although the latter did not refer in his review to a Narrative Medicine approach.

In this sense, it seems very opportune to define the boundaries of the Narrative Medicine approach in order to give it a scientific independence and common protocols to implement. Another important topic that emerged from the review is that studies on Narrative Medicine have used this approach both as an intervention and as a tool to collect narrative data. From our point of view, these two different types of Narrative Medicine studies also imply different research goals. The intervention studies included in the present review were focused on the use of Narrative Medicine to collect information on the effectiveness of this approach and on the patient benefits which derived from it. In this sense, the main aim of the intervention studies on Narrative Medicine seems to be that of assessing the efficacy of the use of the approach on patients: data collected would be evidence to spur health staff into using it in their daily practice.

If instead we look at studies which underline Narrative Medicine as an instrument for collecting narrative data we see that these aim to stress the importance of the approach for providing qualitative information on the patient's experience of their illness experience to the health staff.

Thus, data collected would provide practical knowledge to take into account in medical practice.

For instance, physicians caring for women who have undergone transplantation should take into consideration that the discovery of the pregnancy is a very delicate moment in their patients' lives and calls for particular attention to, and organization of, healthcare [27]. Greenhalgh and Hurwitz [5], in fact, argued that "narratives offer a method for addressing existential qualities such as inner

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1 hurt, despair, hope, grief, and moral pain which frequently accompany, and may even constitute,
2 people's illnesses" (p. 318).

3 Considering the procedural limitations of reviewed works, the systematic review has shown that
4 studies have engaged the participation of very different samples for size, patients' main age and
5 type of disease. Considering that every disease has its own individual care path and thus is
6 associated with different physical and psychological experiences, depending on patients'
7 characteristics, it seems opportune to take into account the individual disease in the design planning
8 of Narrative Medicine studies with patients. More information on the type of disease and the sample
9 characteristics should be addressed in future studies.

10 Furthermore, intervention studies overall did not report the integration of control groups in their
11 research design. Except for the randomized trial conducted by Cepeda and colleagues [20], all other
12 studies have investigated the role of Narrative Medicine interventions without comparing them with
13 other interventions or control groups. Also, the nature of the intervention greatly varies. Participants
14 in the studies included in the review have performed telephone autobiographical interviews,
15 manuscript editing, theatre performance, drawing, storytelling groups, and twenty-minute narrative
16 sections.

17 Another limitation is the high variety of data coding of narratives collected in the research studies
18 using Narrative Medicine as a tool for collecting patients' experiences. In one case [26] researches
19 implemented a software of textual analysis assessing the emerged clusters of narratives, in the other
20 studies authors preferred to conduct a thematic, plot, or narrative analysis.

21 To conclude, it seems very suitable to define the boundaries of the Narrative Medicine approach
22 when it is used in research with patients in order to give it scientific independence and common
23 protocols to implement. Thus, intervention programs should be compliant with the theoretical
24 framework, as well as the analysis of patients' experiences collecting through a Narrative Medicine
25 approach. Starting from the copious scientific literature on the topic, researchers should find a

1 common methodology and a shared procedure which will give the opportunity to replicate the study
2 in other contexts and with patients suffering from different diseases [32, 33, 34].

3 In 2014 in Rome, a committee of international experts in the field participated to a Consensus
4 Conference on recommendations for the implementation of Narrative Medicine in clinical practice
5 [35]. The committee declared to define Narrative Medicine as a methodology of clinical
6 intervention based on a specific communicative competence. Narrative has also defined as a
7 fundamental tool to acquire, comprehend and integrate the different points of view of all the
8 participants having a role in the illness experience. In this sense, the main aim of the Narrative
9 Medicine approach would be that of co-construct a shared and personalized care path [35]. Authors
10 agree with the cited definition and recognize the important role of considering Narrative Medicine
11 as a tool for clinicians daily practice and communication with their patients. In this sense, Narrative
12 Medicine has to be considered as a part of a new broader culture change stressing the importance of
13 a humanization of the care and a personalized Medicine [36,37] tailored and constructed on the
14 individual experience [38], story and needs of every patient.

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22 authors contributed to manuscript preparation and approved the final version.

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Declaration of competing interests

We have read and understood BMJ policy on declaration of interests and declare that we have no competing interests

Data sharing statement

No additional data available

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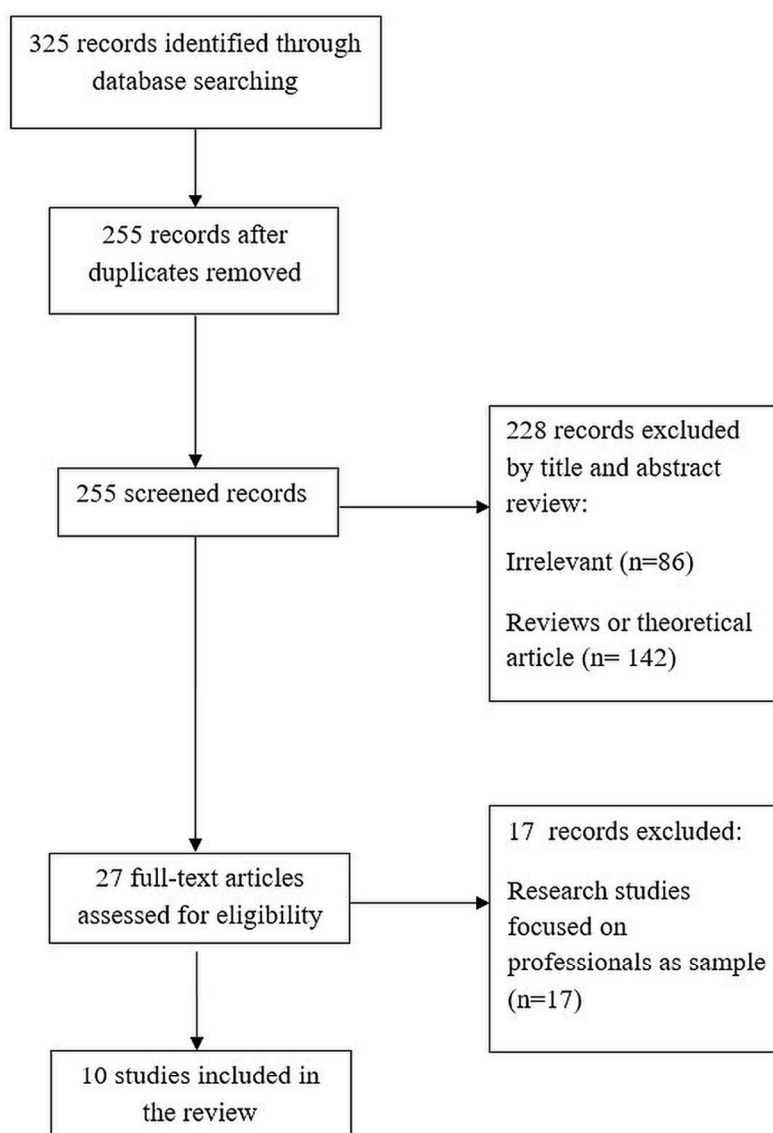


Figure 1. Research studies on patients and caregivers conducted with the Narrative Medicine approach (2005/2015)
116x149mm (300 x 300 DPI)



PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	3-5
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	7
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	NR
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	6
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	6
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	6
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	6
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	NR
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	NR
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I ²) for each meta-analysis.	NR



PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	NR
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	NR
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	7
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	7-14
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	7-14
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	7-14
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	NR
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	NR
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	NR
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	15-17
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	17
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	17
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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