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a systematic review
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Consequences of clinical case management for caregivers: a systematic review

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Running head : Case management and caregivers
Abstract

Background: Informal caregivers are deeply involved in the case management process. However, little is known about the consequences of such programs for informal caregivers. This systematic literature review, reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Statement, addressed the consequences of clinical case management programs, whether positive or negative, for caregivers of frail elderly persons or persons with dementia.

Methods: We systematically identified and analyzed published randomized trials and quasi-experimental studies comparing case management programs to usual care which discussed outcomes concerning caregivers.

Results: Sixteen studies were identified and 12 were included after quality assessment. Seven identified at least one positive result for caregivers, and no negative effect of case management has been found. Characteristics associated with positive results for caregivers were a high intensity of case management and programs specifically addressed to dementia patients.

Conclusion: Despite the numerous methodological challenges in the assessment of such complex social interventions, our results show that case management programs can be beneficial for caregivers of dementia patients, and that positive results for patients are achieved without increasing caregivers' burden.

Key words
Systematic review, older persons, clinical case management, caregiver, persons with dementia.
Introduction

Informal caregivers play a critical role in long-term home care and have been described as the “backbone” of the long-term care workforce (Wolff and Kasper, 2006). Because the stresses of modern society make the traditional mechanisms of providing informal care more difficult, public policy-makers have sought to find efficient ways to support caregivers (Bodsky et al., 2003). A 2002 meta-analysis (Sörensen et al., 2002), pooling a wide range of interventions with caregivers, found that appropriate interventions can lead to significant improvements in caregiver burden, level of depression, and subjective well-being, with a small to moderate effect size. Psychoeducational and psychotherapeutic interventions showed more consistent results, whereas intervention effects tended to be smaller for caregivers of dementia patients. A 2008 systematic review of literature on dementia caregivers (Parker et al.) found only weak evidence that caregivers benefit from such support. Multidisciplinary case management, as analyzed in the three trials examined by the study, seemed to decrease depression scores in caregivers, but this result was not statistically significant. Paradoxically, interventions such as respite care increased caregiver burden. A recent review (Berthelsen et al., 2015) considered effects of case management addressed to informal caregiver of older adults, both on patients and caregivers. Because of a restrictive search algorithm, with a mandatory focus on health information or shared decision making, and methodological limitations (inconsistency in the definition of case management and of caregiver), this review don’t provide reliable results concerning caregivers.

However, case management, which can be defined as a “targeted, community-based and pro-active approach to care that involves case-finding, assessment, care planning, and care coordination,” always involves both the patient and the caregivers (Ross et al., 2011). This is especially true considering Applebaum & Austin (1990) definition of case management,
characterized by three factors: intensity of involvement, breadth of services spanned, and long-term commitment.

Thus, there are two reasons to monitor consequences of case management on caregivers. First, the caregiver can be seen as a part of the caring system, and caregiver support can enhance quality of the care the patient receives. Second, personal interests of the caregiver should be taken into account. Consequences could be beneficial or detrimental, as case management may either reduce or increase caregiver burden.

In keeping with this perspective, the present review has studied the full range of consequences of case management for caregivers of frail elderly persons or persons with dementia. A systematic retrieval of randomized clinical trials and quasi-experimental studies comparing case management and usual care was conducted, in order to answer the following questions: 1) How does case management affect caregivers? 2) What aspects of the case management program could explain the variations in consequences for caregivers? 3) What patient or caregiver characteristics could explain variations in consequences for caregivers? 4) Is case management efficiency for care recipients associated with a positive or negative effect on caregivers?

Methods

The systematic review presented here was undertaken by one of the authors and was not preregistered as a protocol. This report meets the criteria of the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) statement for systematic reviews (Moher et al., 2009).

The present study focuses on primary caregivers of elderly patients with loss of autonomy or persons with dementia without age limit. Patients included in the retrieved studies were considered “elderly” if their average age was more than 75 years. The definition of caregiver was the one chosen by the researchers in each trial, which did not necessarily include a family link or a shared home.
In order to make valid comparisons among the explored interventions, only studies in which “a designated care manager combines the planning and coordination with a therapeutic, supportive role” (Challis et al., 2002), under a model referred to as clinical case management (Kanter, 1989; Surber, 1994) or as intensive care management (Challis et al., 2002), were included. Only studies comparing the specified case management program approach to standard care without any form of case management were included. Quasi-experimental studies were retained because those studies can have a high external validity (Koch et al., 2012). Case management programs that provided only distance services or were limited to education were excluded, as were short term discharge intervention (i.e. transitional care) lasting less than 3 months. All relevant studies published in peer-reviewed journals were considered, regardless of publication date and language. Concerning time-limited interventions, only the outcome assessment at the end of the program was retained; post hoc studies on part of a sample were discarded.

We began our electronic search on the Scopus Database, which includes all MEDLINE articles and indexes 18,500 peer-reviewed journals, including human sciences and non-English journals (http://www.info.sciverse.com/scopus/scopus-in-detail/facts). We developed our search algorithm from a first non-systematic literature review, using index mapping (Strech et al., 2008). We searched in title, abstract, and keywords for the terms “case management” or “care management” or “disease management” or “guided care” or “family intervention” or “integrated care,” in association with “controlled study” or “comparative study” or “prospective experimental design,” associated with “caregiver” in the text. This search, conducted in May 2016, was limited to articles, and articles indexed with “child” as a keyword were excluded. Complementary searches were performed in additional databases, that is, EMBASE, ISI Web of Knowledge, the Cochrane Central Register of Controlled Trials, PsycINFO, CSA Social Services Abstract, and FRANCIS.
All articles were screened by title, and all abstracts of possibly relevant articles were read. Full texts of potentially relevant studies were then retrieved and checked against inclusion and exclusion criteria. Reference lists of selected articles were checked in order to identify other relevant studies. No authors were contacted.

The methodological quality of the randomized studies was systematically assessed for each outcome using the Cochrane Collaboration tool for assessing risk of bias in randomized trials (Higgins et al., 2011). This tool focuses on internal validity and suggests judging the risk of bias in five domains: selection, performance, detection, attrition, and reporting. We added a systematic examination of implementation (Boutron et al., 2008) and contamination biases, because of their relevance to the evaluation of socially complex services (Wolff et al., 2000). Results were then summarized as low, acceptable, high, or unclear.

The potential biases resulting from the absence of blinding differed according to the outcome considered. We maintained that there was a high risk of performance bias on the outcome “satisfaction with care” as caregivers who felt they had “won” the randomization process, or get the “best” service in quasi-experimental studies, might well feel more satisfied. In addition, we saw a high risk of detection bias on the outcome “declared hours of caring,” especially when no in-depth research interview were undertaken, since a case manager’s assessment could influence the caregiver’s awareness of his/her involvement in everyday care. We considered the performance bias on the caregiver’s quality of life to be acceptable, as it has been demonstrated that the Hawthorne effect (i.e., the bias linked with attention and multiple assessments), is not large enough to significantly affect quality of life for caregivers of dementia patients (Mc Carney et al., 2007). By extension, we considered the potential performance bias linked with the absence of blinding on caregiver burden and depression, for both dementia and non-dementia patients, to be acceptable. Non-randomized studies were classified according to the same criteria, taking into account their inherent risk of selection bias. Internal validity assessment was conducted by two reviewers independently, both experienced researchers in the area of case management. Inconsistencies were
resolved by discussion. Results of studies with low internal validity were excluded from further analysis. In included studies, outcomes with high risk of bias were not considered. 

As quality cannot be reduced to internal validity, external validity was assessed as well (Verhagen et al., 2001). In keeping with Wolff's recommendation on socially complex services evaluation (Wolff et al., 2000), we defined two important domains: the representativeness of the research sample, and the accuracy of the description of the interventions, that allows replication of the intervention in similar contexts. External validity was classified as low, acceptable, high, or unclear.

In order to identify elements that might explain differences in outcomes, we systematically collected program and population characteristics. Program descriptions were analyzed in order to assess program intensity and level of integration. The simplest way to evaluate case management intensity is to consider the case manager’s case load (the greater the case load, the lower the intensity) (Somme et al., 2012). To refine this approach, we also considered the frequency of home visits, the range of services (Pacala et al., 1995), and the complexity of the needs of the target population (Challis 2002). We estimated level of integration on the basis of case managers' links with primary care practice, service providers, acute-care facilities, and long-term-care facilities (Kodner, 2002). Intensity and integration were estimated as low, moderate, high, or unclear. As no consensually validated tool exists to assess external validity, case management intensity, and case management integration, these assessments were made by the two reviewers through continuous discussions to achieve agreement.

Finally, in order to identify elements that might explain differences in outcomes, we systematically collected program and population characteristics. Program descriptions were analyzed in order to assess program intensity and level of integration. Relevant characteristics of the caregiver population were determined – in particular, the proportion of women, and the proportion of spouses, since those characteristics are known to influence caregiver burden (Carretero et al., 2009). We also collected statistically significant results
concerning patients, in order to assess the relationship between patient and caregiver outcomes.

A vote-count review was performed on caregiver outcomes, summarizing those reflecting burden (or strain or distress), satisfaction, health-related quality of life (or psychological health), depression, and social support. Sub-group analyses were carried out in terms of patient characteristics (persons with dementia versus elderly persons) and patient results, caregiver characteristics, and intensity and integration levels for case management.

Results

Selection of Studies

Our bibliographic search in Scopus identified 481 articles. The selection process (Figure 1) led us to include 16 studies (Table 1). Frequently assessed outcomes were caregiver burden (13 studies), caregiver health-related quality of life or psychological health (8 studies), caregiver depression (4 studies), satisfaction with care (3 studies), or social support (3 studies). Other outcomes considered by at least one study were caring hours, caregiver sense of competence, personal well-being, family functioning, desire to institutionalize, productivity loss and loneliness.

Eleven randomized controlled trial studies and five quasi-experimental studies were analyzed. No randomized trial was blinded for participants, which makes sense, as such blinding would be difficult, if not impossible, for case management. Assessment was blinded in 11 studies, non-blinded in 5, and unclear in one. Three of the five quasi-experimental studies were considered to have low internal validity, mostly because of significant differences between experimental and control groups at inclusion. One randomized controlled trial with only one caregiver outcome, declared hours of caring, was considered to have low internal validity for this outcome because of a detection bias. These four studies were excluded from further analysis. Reviewers agreed on conclusions concerning internal
validity for all randomized trials, but discussions were necessary for the two quasi-experimental studies that have finally been accepted.

There were no studies with high internal validity. The 12 remaining studies were judged to have unclear (2 studies) or acceptable (10 studies) internal validity, in spite of moderate risks of performance, detection, implementation, or contamination bias. Five studies had low external validity, mainly because they lacked a systematic recruitment strategy. Table 2 summarizes the results concerning the internal and external validity of each study, and specifies the kinds of bias that could affect the results. We included two studies published by Chien and Lee (2008,2011) despite some questions about these reports: they describe two very similar trials without discussion on the additional value of the second study.

Outcomes for Caregivers

Seven of the 12 studies found at least one valid positive outcome for caregivers [Table 3]. Four of the 11 studies assessing caregiver burden showed a reduction. Four of the 7 studies assessing quality of life for caregivers yielded positive results; one of 3 on caregiver social support showed positive results. The 4 studies that assessed depression in caregivers found no significant differences at the end of the intervention. Callahan (2006) found a significant result on depression score 6 months after the end of the intervention. No studies demonstrate any negative effects of case management on caregivers.

Pre-specified Sub-group Analysis

In further analyses we tried to understand which aspects of a case management program could explain variations in the program's efficacy for caregivers. As it was not possible to control for all confounding factors, especially the variability of the context, the further analyses should be regarded as exploratory.
To analyze intensity and integration influence, we separate here two different case management models compared with control group by McNeil: the “intensive” one, and the “linkage model”.

Patient profiles are important determinants of efficacy. As Table 4 shows, none of the programs aimed at elderly patients showed positive outcomes for caregivers. Selection criteria were different (table1) according to the studies, but all these, except in Leung study (2004), can be qualified of “frail”, because of a loss of autonomy or a high risk of using health service resource. Six of the ten programs targeted to dementia patients found positive outcomes for caregivers, the other having both a low or moderate integration and a moderate or unclear intensity level. The two studies aimed at mild-dementia patients only demonstrated no positive outcome. Five of the six studies with a high intensity level of case management achieved positive outcomes for caregivers whereas only one of the five studies with a moderate level of intensity achieved positive outcomes for caregivers. The level of integration of a case management program, patient age, or caregiver characteristics did not seem to figure highly in program efficacy.

It is important to emphasize that these results are based only on caregiver outcomes. In terms of patient outcomes, 9 of the 12 studies reported at least one positive outcome for patients, such as delaying institutionalization, reducing hospitalization, enhancing quality of life, or reducing behavioral troubles. Some programs may yield significant effects for patients, especially frail elderly, without benefit for caregivers. Parsons’ study (2012) demonstrates the possibility of benefit in the form of delayed institutionalization, which was not mediated by a reduction of caregiver burden.

**Discussion**

Evidence concerning socially complex services is difficult to gather (Wolff, 2000). Because of the heterogeneity of study designs, interventions, contexts, and outcomes considered, we did not attempt to pool the results in a meta-analysis. We believe that this overview obtained
through careful individual assessment of study quality, program, and outcomes can be more informative than any summary measure obtained via global pooling of different interventions. Although some uncertainty remained, due to the impossibility of conducting blinded trials, this systematic review allows us to conclude that some case management programs aimed at dementia patients can have positive outcomes for caregivers, in terms of reduction of caregiver burden, enhanced quality of life, or greater social support, but not on depression. Caregivers of patients with mild dementia may not be the persons who benefit most from case management programs (Jansen et al., 2011). There is no evidence that case management programs may have positive outcomes for caregivers of frail elderly, but such programs can reduce hospitalization (Leung et al., 2004; Béland et al., 2006) or delay institutionalization (Parson et al., 2012) without increasing caregiver burden.

Our systematic review has several limitations. In addition to the usual publication bias, identification of all the relevant studies has proven challenging, since case management programs can be described by different terms. Despite the care we took in building a systematic search algorithm, it is possible that some relevant studies were not identified. Ultimately, however, any real weakness in this systematic review lies in the quality of the studies themselves, all of which were based on subjective outcomes which could have been biased by the absence of blinding. Another potentially important bias concerns implementation, as most of the studies reported no implementation results. Three studies attributed disappointing results to inadequate program implementation (Vickrey et al., 2006; Jansen et al., 2011; Fox et al., 2000). Contamination may also have occurred, especially in trials with individualized randomization: good practices implemented by case managers could have also benefitted the control group, if the same professionals were involved in both groups. Whereas the implementation and contamination bias may have reduced the results, the non-blinded design might have increased them.

It has proved difficult to identify aspects of case management program and patients selection associated with better results. This is mainly due to an insufficient reporting, and to the
heterogeneity of studies. Intensity and integration levels were especially difficult to determine. When MacNeil compares with control group two different models of case management, named “intensive” and “linkage”, it happens that the case load is similar in the two models (more important in the linkage model, though non significant), the intensive model referring in fact to a more integrated model, more efficient on caregiver psychological health. This stresses the necessity of a complete description of implemented interventions.

The value of quasi-experimental design in evaluation of case management programs is somewhat controversial (Koch et al., 2012; Wolff, 2000). Hébert (2000) was able to provide interesting data on the strength of an extensive (4 year) follow-up of an “actually implemented” program, and it emphasizes the time required to implement a program before assessing its efficacy. Quasi-experimental studies can avoid contamination bias, when different territories are compared, and their implementation, even when only partial, corresponds to “real-life” implementation. We believe that the performance bias linked with the absence of patient blinding could be decreased by the absence of randomization, as patients would not have felt they had won something others had not. However, the detection bias is increased by the absence of a blinding assessment, usually infeasible in such studies [8]. The main problem, as observed in four such studies (Morales-Asencio et al., 2008; Hébert et al., 2010, Janse et al., 2014, McNeil et al., 2015) is the difficulty to obtain two similar groups that could allow comparison. Despite our initial choice of including quasi-experimental study, three of five were finally not included because of significant differences in the compared groups, added with at least one other identified bias, and for the two included trial there was initially a disagreement between the expert. Challis (2002) design, with similar group obtained by matching patient pairs, and MacNeil (2015) statistical analysis using propensity score, seems valuable ways to enhance internal validity of quasi-experimental studies. Further studies could consider cluster randomization, eventually with stepped wedge design to model the effect of time (Hemming, 2015).
Lastly, all studies included only one caregiver per patient, with different definition and often little details provided; so the effect of case management on “second line” caregivers or according to the characteristics of caregivers (spouses versus relatives versus friends) can’t be specified.

Despite these limitations, we believe we have been able to gather valuable evidence on the potential benefits for caregivers of case management. Our rigorous selection of studies, based on a clear definition of case management, allow confidence in the conclusions drawn. Positive results have been obtained in very different cultural and organizational contexts, in Asia, Europe, and America. Despite the limited external applicability of some individual studies, this review provides a good representation of different contexts overall. Of course, the literature cannot provide evidence that every case management program will work everywhere, nor that any given single model will work in every context. In contrast to drug trials, case management is an intervention that cannot be completely standardized (Wolff, 2000), and researchers studying case management have to make compromises between internal and external validity (Godwin et al., 2003). So our review can only stress the possibility of implementing efficient case management programs for caregivers, and highlight the characteristics that seem to be associated with better outcomes. Until now, such programs have been nationally implemented in very different contexts, for example in Japan (Matsuda and Yamamoto, 2001), France (Somme et al, 2009), or Québec, Canada (Vedel et al, 2011).

No study showed any effect of a program on caregiver depression. That said, the relevance of such an outcome for a case management program can be questioned: if depression is envisaged as a systemic disease, involving the mind, brain and body (Gold and Charney, 2002) such intervention could address only social, external components of depression. The choice of assessment instruments for depression can also be questioned (Snaith 1993; Roman and Callen, 2008), the scales chosen to rate caregiver burden. The construction of scales is “necessarily influenced by a variety of value judgements” (Janse 2014). Burden
scales that focus on the negative impacts of caregiving have been criticized, because that restriction prevents researchers from assessing sources of satisfaction in caregiving (Nolan 2006). Other scales that include both negative and positive impacts of caregiving could be more relevant to assessment of the global consequences of caregiving (McKee et al., 2003). The finding that studies aimed at frail elderly can yield positive results for patients without significantly affecting caregivers is in itself valuable. As such programs involve both caregivers and patients, it seems good practice to evaluate the respective effects on both. Five studies raise questions about possible harmful effects of case management. In the Hébert study (2010), the burden increase was significantly greater in the intervention group. One hypothesis is that patient empowerment was achieved at the expense of caregiver burden. Two studies (Leung et al., 2004; Vickrey et al., 2006) found a non-significant trend in favor of an increase of caregiving hours in the intervention group, which conflicts with Challis (2001) results. It is possible that in some contexts case managers might ask caregivers to perform new caregiving tasks. Lam (2010) found a slight negative effect at 12 months on caregivers’ psychological health (our review focused on the effect at the end of the intervention at 4 months), and assumed that domestic helpers’ lack of experience with dementia patients, and the cost of day care facilities, may have in fact been burdensome to some family caregivers. Finally, Janse (2014) complementary study on caregiver satisfaction demonstrates the possibility of counterproductive effect of case management due to information overload or decrease clarity on “who do what” for caregiver. These results, although a minority, emphasize the necessity of considering caregiver outcomes in all case management intervention, at least as a possible side effect.

Conclusion

Our results show reasonable evidence that clinical case management can lead to positive outcomes for caregivers of dementia patients, and that positive outcomes for frail elderly
patients can be obtained without increasing caregiver burden. Further research is needed to
determine which characteristics of a program, in addition to intensity, could support the
program's efficacy, and to determine which caregivers could benefit most from those
programs. Even though researchers face numerous methodological challenges in building
valid trials, evidence can be strengthened via accurate description of the context, of the
intervention itself, and of its actual implementation, as well as through a careful choice of trial
design and outcomes.

Figure 1 : Flowchart of Selection of Studies

Declarations

- Competing interests

The authors declare that they have no competing interests.

Key points

- Informal caregivers are deeply involved in the case management process
- Case management programs can achieve positive results for caregivers, especially
  intensive programs addressed to persons with dementia.
- Positive results for older patients, such as delaying institutionalization and reducing
  hospitalization, occur without impacting caregivers’ outcome.
References


doi: 10.1016/j.jamda.2015.06.011.


Scopus systematic research
481 citations identified

342 citations excluded based on information in title

139 abstracts retrieved

83 citations excluded based on abstract screening
The intervention is not a clinical case management program (n = 47)
No outcomes on caregiver (n = 18)
Case managers in the 2 groups (n = 9)
The intervention concerns younger populations (n = 5)
No comparative study (n = 4)

56 full text articles assessed for eligibility

38 articles excluded after full text review
No outcomes on caregiver (n = 18)
The intervention is not a clinical case management program (n = 15)
Case managers in the 2 groups (n = 5)
Post-hoc study on part of a sample (n = 1)

17 articles retained, corresponding to 16 included studies

Figure 1: Flowchart of Selection of Studies
<table>
<thead>
<tr>
<th>Author, Year</th>
<th>Target Population (patient)</th>
<th>Patient Mean Age</th>
<th>Number of Caregivers</th>
<th>Caregiver Mean Age</th>
<th>% women</th>
<th>% spouse</th>
</tr>
</thead>
<tbody>
<tr>
<td>Newcomer 1999</td>
<td>&gt;65 years old, Diagnosis of dementia</td>
<td>78</td>
<td>5307</td>
<td>63</td>
<td>60</td>
<td>50</td>
</tr>
<tr>
<td>Challis 2002</td>
<td>dementia, significant unmet needs, risk of institutionalisation</td>
<td>80</td>
<td>86</td>
<td>59</td>
<td>Unknown</td>
<td>0.2</td>
</tr>
<tr>
<td>Leung 2004</td>
<td>&gt;60 years old, discharged from a rehabilitative hospital</td>
<td>75</td>
<td>Unknown (260 patients)</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
</tr>
<tr>
<td>Béland 2006</td>
<td>&gt;64 years old, at least moderate disability</td>
<td>82</td>
<td>1309</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
</tr>
<tr>
<td>Vickrey 2006</td>
<td>&gt;65 years old, diagnosis of dementia</td>
<td>80</td>
<td>408</td>
<td>66</td>
<td>69</td>
<td>56</td>
</tr>
<tr>
<td>Callahan 2006</td>
<td>&gt;65 years old, diagnosis of dementia</td>
<td>77.5</td>
<td>153</td>
<td>61</td>
<td>89</td>
<td>44</td>
</tr>
<tr>
<td>Chien 2008</td>
<td>Diagnosis of Alzheimer disease</td>
<td>68</td>
<td>88</td>
<td>44</td>
<td>64</td>
<td>32</td>
</tr>
<tr>
<td>Morales 2008</td>
<td>Terminally ill or dependant patient who require daily assistance</td>
<td>76</td>
<td>205</td>
<td>57</td>
<td>79</td>
<td>25</td>
</tr>
<tr>
<td>Wolff 2009</td>
<td>&gt;65 years old; high risk of using health service resource</td>
<td>78</td>
<td>308</td>
<td>61</td>
<td>70.4</td>
<td>48</td>
</tr>
<tr>
<td>Hébert 2009</td>
<td>&gt;=75, risk of functional decline</td>
<td>83</td>
<td>1166*</td>
<td>Unknown</td>
<td>75*</td>
<td>35*</td>
</tr>
<tr>
<td>Lam 2010</td>
<td>&gt;65 years old, mild dementia</td>
<td>78</td>
<td>102</td>
<td>Unknown</td>
<td>75</td>
<td>30</td>
</tr>
<tr>
<td>Chien 2011</td>
<td>Alzheimer disease, mild or moderate stage</td>
<td>67.8</td>
<td>92</td>
<td>45</td>
<td>66</td>
<td>27</td>
</tr>
<tr>
<td>Jansen 2011</td>
<td>&gt;65 + diabetes or 75 years old, and dementia</td>
<td>81</td>
<td>99</td>
<td>62</td>
<td>69</td>
<td>40</td>
</tr>
<tr>
<td>Parsons 2012</td>
<td>&gt;65 years old (55 for Maoris); high institutionalisation risk</td>
<td>81</td>
<td>115</td>
<td>67</td>
<td>60</td>
<td>50</td>
</tr>
</tbody>
</table>
Table 2: Quality assessment of the studies, detailed by outcome

<table>
<thead>
<tr>
<th>Author, Journal, Year</th>
<th>Internal Validity*</th>
<th>External Validity</th>
</tr>
</thead>
<tbody>
<tr>
<td>Newcomer Health services Research 1999</td>
<td>Non-blinded RCT High risk of detection bias on the total hours of informal care, with a possible under-estimation in control group Implementation bias threw the different sites <strong>Unclear</strong></td>
<td>Recruitment based on voluntary application and little description of CM tasks <strong>Low</strong></td>
</tr>
<tr>
<td>Challis Int J of Geriatric Psychiatry 2002</td>
<td>Quasi-experimental design Matched pairs, allowing similar group Possible detection bias linked with a non-blinded assessment High risk of performance bias on the outcome “satisfaction with care” High risk of detection bias on the total hours of informal care <strong>Acceptable</strong></td>
<td>Systematic recruitment; unclear description of the intervention <strong>Moderate</strong></td>
</tr>
<tr>
<td>Leung J of applied gerontology 2004</td>
<td>Non-blinded RCT Assessment blinding unclear High risk of detection bias on the total hours of informal care <strong>Low</strong> concerning the total hours of informal care</td>
<td>No systematic recruitment No description of the caregiver population <strong>Low</strong></td>
</tr>
<tr>
<td>Béland J of Gerontology 2006</td>
<td>Non-blinded RCT High risk of performance bias on the outcome satisfaction with care Unclear risk of contamination <strong>Acceptable</strong></td>
<td>No systematic recruitment <strong>Moderate</strong></td>
</tr>
<tr>
<td>Vickrey Annals of internal medicine 2006</td>
<td>Non-blinded RCT No validated tool for caregiver confidence and sense of mastery Limited implementation <strong>Acceptable</strong></td>
<td>High</td>
</tr>
<tr>
<td>Callahan JAMA 2006</td>
<td>Non-blinded RCT <strong>Acceptable</strong></td>
<td>High refusal rate for a complete diagnosis process <strong>Moderate</strong></td>
</tr>
<tr>
<td>Morales BMC Health Services research 2008</td>
<td>Non-blinded quasi-experimental study Base-line difference in functional capacity Progressive implementation, implying a contamination bias</td>
<td>Systematic sampling, but unclear description of the intensity of the intervention <strong>Moderate</strong></td>
</tr>
</tbody>
</table>
Table 3: Caregiver outcomes with acceptable or unclear internal validity detailed by study.

<table>
<thead>
<tr>
<th>Publication</th>
<th>Burden</th>
<th>Depression</th>
<th>Health Related Quality of life</th>
<th>Social support</th>
<th>Other caregiver outcomes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Newcomer 1999</td>
<td>Adapted from Zarit Carer Burden Interview</td>
<td>Geriatric depression Scale (short form)</td>
<td>Not assessed</td>
<td>Not assessed</td>
<td>No</td>
</tr>
<tr>
<td>Challis 2002</td>
<td>Adapted from the Social Behaviour Assessment Schedule Caregiver strain (Malaise Scale)</td>
<td>Not assessed</td>
<td>Not assessed</td>
<td>Not assessed</td>
<td>No</td>
</tr>
<tr>
<td>Béland 2006</td>
<td>Zarit Carer Burden Interview</td>
<td>Not assessed</td>
<td>Not assessed</td>
<td>Not assessed</td>
<td>No</td>
</tr>
<tr>
<td>Vickrey 2006</td>
<td>Not assessed</td>
<td>Not assessed</td>
<td>Euroqol 5D</td>
<td>Medical Outcome Study Social Support</td>
<td>Confidence in caregiving; Caregiving Mastery</td>
</tr>
<tr>
<td>Callahan 2006</td>
<td>Caregiver Neuropsychiatric inventory (caregiver distress)</td>
<td>Patient Health Questionnaire 9</td>
<td>Not assessed</td>
<td>Not assessed</td>
<td>No</td>
</tr>
<tr>
<td>Chien 2008</td>
<td>Family Caregiver Burden Inventory</td>
<td>Not assessed</td>
<td>World Health Organization Quality of Life Scale</td>
<td>Six-item Social Support Questionnaire</td>
<td>No</td>
</tr>
<tr>
<td>Wolff 2009</td>
<td>Modified Caregiver Strain Index</td>
<td>Center for Epidemiologic Studies Depression Scale</td>
<td>Not assessed</td>
<td>Not assessed</td>
<td>Productivity loss</td>
</tr>
<tr>
<td>Lam. 2010</td>
<td>Zarit Carer Burden Interview</td>
<td>Not assessed</td>
<td>General Health Questionnaire 30; Personal Well-Being Index</td>
<td>Not assessed</td>
<td>No</td>
</tr>
<tr>
<td>Chien 2011</td>
<td>Family Caregiver Burden Inventory</td>
<td>Not assessed</td>
<td>World Health Organization Quality of Life Scale</td>
<td>Six-item Social Support Questionnaire</td>
<td>No</td>
</tr>
<tr>
<td>Jansen 2011</td>
<td>Self Perceived Pressure by Informal Carer</td>
<td>Center for Epidemiologic Studies Depression Scale</td>
<td>Medical Outcome Study 36 items Short form</td>
<td>Not assessed</td>
<td>Caregiver sense of competence (Sense of Competence Questionnaire)</td>
</tr>
<tr>
<td>Parsons 2012</td>
<td>Caregiver Reaction Assessment</td>
<td>Not assessed</td>
<td>Medical Outcome Study 36 items Short form</td>
<td>Not assessed</td>
<td>No</td>
</tr>
<tr>
<td>MacNeil 2015</td>
<td>CarerQOL</td>
<td>Not assessed</td>
<td>Euroqol 5D</td>
<td>Not assessed</td>
<td>Short sense of competence scale Loneliness scale</td>
</tr>
<tr>
<td>Patient Profile</td>
<td>Duration of the Case Management Program (in months)</td>
<td>Intensity</td>
<td>Integration</td>
<td>Positive Outcomes Concerning Caregivers</td>
<td>Positive Outcomes Concerning Patients</td>
</tr>
<tr>
<td>----------------</td>
<td>-----------------------------------------------------</td>
<td>-----------</td>
<td>-------------</td>
<td>----------------------------------------</td>
<td>---------------------------------------</td>
</tr>
<tr>
<td>Challis 2002</td>
<td>dementia 12</td>
<td>high</td>
<td>high</td>
<td>lower burden</td>
<td>less institutionalisation improvement in social contacts, need reduction, level of risk reduction</td>
</tr>
<tr>
<td>Callahan 2006</td>
<td>dementia 12</td>
<td>high</td>
<td>low</td>
<td>lower burden</td>
<td>lower Neuro-psychiatric inventory</td>
</tr>
<tr>
<td>Chien 2008</td>
<td>dementia 6</td>
<td>high</td>
<td>moderate</td>
<td>lower burden, better QoL</td>
<td>less institutionalisation; decrease of symptom severity</td>
</tr>
<tr>
<td>Chien 2011</td>
<td>dementia 6</td>
<td>high</td>
<td>moderate</td>
<td>lower burden, better QoL</td>
<td>less institutionalisation</td>
</tr>
<tr>
<td>MacNeil2015 Intensive CM</td>
<td>dementia 24</td>
<td>Unclear</td>
<td>High</td>
<td>better QoL</td>
<td>no</td>
</tr>
<tr>
<td>Vickrey 2006</td>
<td>dementia 18</td>
<td>moderate</td>
<td>moderate</td>
<td>better social support and confidence in caregiving</td>
<td>higher care quality; higher quality of life; better social support; fewer unmet needs</td>
</tr>
<tr>
<td>Newcomer 1999</td>
<td>dementia 36</td>
<td>unclear</td>
<td>low</td>
<td>no</td>
<td>fewer unmet needs</td>
</tr>
<tr>
<td>MacNeil2015 Linkage model</td>
<td>dementia 24</td>
<td>Unclear</td>
<td>low</td>
<td>no</td>
<td>no</td>
</tr>
<tr>
<td>Leung 2004</td>
<td>elderly 6</td>
<td>moderate</td>
<td>high</td>
<td>no</td>
<td>less hospitalisation</td>
</tr>
<tr>
<td>Béland 2006</td>
<td>frail elderly 22</td>
<td>high</td>
<td>high</td>
<td>no</td>
<td>less hospitalisation</td>
</tr>
<tr>
<td>Wolff 2009</td>
<td>frail elderly 18</td>
<td>moderate</td>
<td>moderate</td>
<td>no</td>
<td>Reduction in the use of home health care</td>
</tr>
<tr>
<td>Lam 2010</td>
<td>mild dementia 4</td>
<td>moderate</td>
<td>moderate</td>
<td>no</td>
<td>no positive results</td>
</tr>
<tr>
<td>Jansen 2011</td>
<td>mild dementia 12</td>
<td>moderate</td>
<td>moderate</td>
<td>no</td>
<td>No effect on quality of life</td>
</tr>
<tr>
<td>Parsons 2012</td>
<td>frail elderly 24</td>
<td>unclear</td>
<td>high</td>
<td>no</td>
<td>fewer residential placement + death</td>
</tr>
</tbody>
</table>

Studies with statistically significant outcomes for caregivers are in the upper part of the table.

QoL: Health related quality of life
The instrument used in each study is listed. Statistically significant results are in bold.
<table>
<thead>
<tr>
<th>Study</th>
<th>Design</th>
<th>Bias and Limitations</th>
</tr>
</thead>
<tbody>
<tr>
<td>Wolff Gerontologist 2009</td>
<td>Non-blinded RCT</td>
<td>High risk of detection bias, as assessment is made by the case managers in the intervention group</td>
</tr>
<tr>
<td>Hébert J of Gerontology 2009</td>
<td>Non-blinded quasi-experimental study</td>
<td>Baseline difference in age (older in experimental group) and caregiver burden (lower in experimental group)</td>
</tr>
<tr>
<td>Chien Psychiatric services 2010</td>
<td>Non-blinded RCT</td>
<td>Unclear contamination, implementation and reporting bias</td>
</tr>
<tr>
<td>Lam Int J of Geriatric Psychiatry 2010</td>
<td>Non-blinded RCT</td>
<td>Unclear contamination and implementation bias</td>
</tr>
<tr>
<td>Chien J of Advanced Nursing 2011</td>
<td>Non-blinded RCT</td>
<td>Unclear implementation and contamination bias</td>
</tr>
<tr>
<td>Jansen Int J of Nursing Studies 2011</td>
<td>Non-blinded RCT</td>
<td>High risk of performance bias for satisfaction with care</td>
</tr>
<tr>
<td>Parsons JAGS 2012</td>
<td>Non-blinded RCT</td>
<td>Systematic recruitment, but insufficient description of the intervention</td>
</tr>
<tr>
<td>Janse</td>
<td>Non-blinded quasi-experimental study</td>
<td></td>
</tr>
<tr>
<td>Study</td>
<td>Bias Description</td>
<td>Recruitment Bias</td>
</tr>
<tr>
<td>--------------</td>
<td>----------------------------------------------------------------------------------</td>
<td>--------------------------</td>
</tr>
</tbody>
</table>
| BMC geriatrics 2014 | Selection bias, with baseline differences in patients (gender and home living)  
Attrition bias, with 40% of the caregiver loss to follow up | No systematic recruitment  
Well implemented intervention | Moderate               |
| MacNeil     | Selection bias corrected by appropriated statistical analysis (propensity score)  
Possible contamination bias | High refusal rate        | Moderate              |

RCT: randomized controlled trial  
CM: case manager
<table>
<thead>
<tr>
<th>Study</th>
<th>Characteristic</th>
<th>Mean</th>
<th>Median</th>
<th>SD</th>
<th>Min</th>
<th>Max</th>
</tr>
</thead>
<tbody>
<tr>
<td>Janse 2014</td>
<td>&gt;75 years old and frail (Groningen frailty indicator)</td>
<td>82</td>
<td>159</td>
<td>63</td>
<td>73</td>
<td>32</td>
</tr>
<tr>
<td>Macneil Vroomen 2015</td>
<td>diagnosis of dementia</td>
<td>80</td>
<td>521</td>
<td>65</td>
<td>67</td>
<td>52</td>
</tr>
</tbody>
</table>

Table 1: Main characteristics of participants, patients and caregivers, in the 16 identified studies.

*extrapolation from data concerning only the first recruitment wave