

The Foundations Framework for developing and reporting new models of care for multimorbidity

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Abbreviations:

CCM: Chronic care model

NICE: The National Institute for Health and Care Excellence

Abstract

Purpose

Multimorbidity is a challenge for health systems globally. New models of care are urgently needed to better manage patients with multimorbidity. However, there is no agreed framework to support development and reporting of models of care for multimorbidity and their evaluation.

Methods

We used a literature search to identify models of care for multimorbidity. We developed a framework to describe these models. We illustrate the application of the framework by identifying the focus and gaps in current models of care, and by describing the evolution of models over time.

Results

The framework describes each model in terms of its theoretical basis and target population (the foundations of the model), and elements of care implemented to deliver the model. We categorised elements of care into three types: (a) clinical focus; (b) organisation of care; (c) support for model delivery. Application of the framework identified a limited use of theory in model development and a strong focus on some patient groups (elderly, high users) more than others (younger patients, deprived populations). We identified changes in elements over time, with a decrease in models implementing home care, and an increase in models offering extended appointments.

Conclusions

By encouraging greater clarity about underpinning theory and target population, and categorising the wide range of potentially important elements of an intervention to improve care for patients with multimorbidity, the framework may have utility in the development and reporting of models of care. This will help ensure the effective development of the currently limited evidence base.

Introduction

Chronic disease is a global health priority,[1 2] and multimorbidity (the co-existence of two or more chronic conditions in a patient)[3] brings additional challenges. Patients with multimorbidity receive more fragmented care and have worse health outcomes, and health systems struggle to effectively address their needs.[3] New ways to deliver care are required to manage the needs of these patients, especially in primary care which often has responsibility for their management.[4-7]

Current progress in improving care for multimorbidity

There are major gaps in the evidence base concerning care for multimorbidity. A recent Cochrane review found only 18 trials evaluating models.[8] Models involved two broad strategies; reorganisation of care delivery through enhanced multidisciplinary teamwork, and patient-oriented education or self-management. The review found limited evidence that the models examined were effective.

Lack of consensus over the description of models for multimorbidity is a significant problem. If science is to drive clinical innovation, we need to build the evidence-base through on-going evaluation and review. However, that process is hampered by incomplete descriptions of models in publications. [9] Without complete, accurate descriptions, researchers cannot replicate studies or identify 'active ingredients'. [10]

Developing a comprehensive framework for developing and reporting models for multimorbidity would provide a common understanding for researchers and clinicians, enable better description of existing and new models, and allow more effective analyses of 'what works for whom' in multimorbidity. This is likely to be critical, given the broad range of potential approaches and different patient populations included under the multimorbidity label. In developing our framework, we adopted Davidson et al.'s definition of a 'model' of care as an:

'overarching design for the provision of a particular type of health care service that is shaped by a theoretical basis, evidence-based practice and defined standards. It consists of defined core elements and principles and has a framework that provides the structure for the implementation and subsequent evaluation of care'.[11]

We had the following aims:

- 1. To identify models of care relevant to the management of multimorbidity.
- 2. To analyse models in terms of their theoretical basis and core elements and develop a framework for the description of models of care.

3. To illustrate the utility of the framework through applying it to explore how models have changed over time.

Methods

Identification of models

We carried out a large scoping review[12] of models of primary care for chronic conditions and multimorbidity. We searched three bibliographic databases (MEDLINE, EMBASE, Cochrane CENTRAL) with blocks of terms for *multimorbidity/chronic conditions*; AND *primary care*; AND *models/frameworks/interventions* (see Appendix for search and model selection details), and supplemented with our knowledge of any additional models that fit our criteria. We did not exclude any model based on study type.

Analysis and framework development

We recorded the underlying theoretical basis of the model, and extracted elements in each published paper. We categorised these elements, formulating a glossary (see Appendix). We also extracted details of the patient populations relevant to the model, and provided a short description. We developed the framework iteratively, with the authors reflecting on the elements of the models as the data were extracted, the structure of those elements and common groupings of elements through group discussion. Drawing from these discussions and the authors' own experience of multimorbidity and health systems research, we proposed a framework.

Application of the framework

We used the framework to summarise the content of current models of care for multimorbidity, highlighting key issues arising from application of our framework to the models. To further illustrate use of the framework, we explored the evolution of models of care over time, comparing components of more recent models from 2010 onwards to earlier models (this gave an approximately even number of models in each period) to illustrate developments over time. We used chi-squared tests to compare the frequency with which elements were included before or after 2010.

Results

Identification of models

Database searching identified 15,880 titles. Following screening and addition of any other relevant models known to the team, we included 39 different models of care (described in 68 papers). The Appendix gives a brief description of each model (Table A1) and their elements (Table A2).

Analysis and framework development

Figure 1 illustrates the framework which arose from our analysis of the models identified and discussion of common groupings and descriptive properties. The *foundations* of the framework are the theoretical basis of the model, and the defined patient population. We categorised the *elements* of the model which are then implemented according to 3 categories: (a) clinical focus; (b) organisation of care delivery; (c) support for model delivery.

[insert Figure 1 – The framework]

The first foundation of a model is its *theoretical basis*. Models of care are designed by agents and generally to achieve a specific goal. For example, they might be designed primarily to improve quality of care and reduce treatment burden for a group of multimorbid patients who previously had to attend multiple additional visits; or, they might be designed primarily to address a system's financial pressures. Models of care are therefore built (at least implicitly) on assumptions of how these goals can be achieved, and these assumptions of the designers are sometimes outlined *explicitly* by reference to a theoretical basis. This does not generally specify in great detail the specific elements used in the model.[13]

The second foundation is the *defined target population*. Despite similarities, chronic conditions vary significantly in their manifestations and treatment indications. Similarly, people with multimorbidity are an extremely heterogeneous population, with many potential subgroups and varying needs. Therefore, a description of the population being targeted by a model is essential.

Elements: Building on the foundations of the theoretical basis and the target population, models have different elements that are actually implemented. Many models of care change the *clinical focus* of care, and changes here will likely be the most visible to the patients. Changes here should align with the theoretical basis of the model (e.g. shared decision-making for 'patient-centered care'), and might reflect the target population (e.g. a focus on mental health).

Models often change the organisation of care delivery, to allow or facilitate the necessary changes in clinical focus. For example, extended appointment times may be needed for shared decision-making, or health coaches might be better suited than physicians to help patients self-manage. Specialist staff may be most efficiently co-located in primary care to prevent multiple visits and unnecessary patient burden.

Adapting the clinical focus and organisation of care delivery is likely to require *support for model delivery* including changes to supporting infrastructure, such as adaptions to workforce, technology, or incentives. For example, funding/payment changes might be necessary to change provider behaviours (e.g. incentivising preventative behaviour), to better integrate care between sectors, or to drive up the quantity of care delivered in the primary care setting.

Application of the framework

Application of our framework to existing models of care for multimorbidity has highlighted the following issues.

Theoretical basis: Of the 39 models identified, only 17 (44%) explicitly named a theoretical basis, and 15 of those (88%) cited the Chronic Care Model (CCM). The CCM describes several basic principles (use of self-management support, clinical information systems, delivery system redesign, decision support, health care organisation, and community resources).[14] In practice, models did not include all principles of the CCM. For example, few examined 'clinical information systems' at all. Other theoretical bases identified included the Patient Centred Care Model,[15] and the Home-based Palliative Care Programme model.[16]

Target populations: Most models (67%, n=26) targeted only those at the highest risk of adverse events (most commonly hospitalisations), and/or the elderly. Only one model focused on those living in socioeconomically deprived areas. Only two models aimed to adapt care for their entire patient population (i.e. taking a population health management approach through a patient-centered medical home), expanding the target group to include more prevention-oriented clinical elements.

Elements: Figure 2 summarises the elements identified in the current models of care. Below we describe elements in the three categories of clinical focus, organisation of care delivery, and support for model delivery. Almost all models (97%, n=38) reported changes across all three categories, but with wide variation in specific elements deployed (see Appendix Table A2).

[insert Figure 2 - Percentage of models in the current literature using each component of the framework]

In terms of *clinical focus*, the majority of models included self-management support (87%), a biopsychosocial approach (79%), a focus on prevention (74%), and attention to polypharmacy (72%). Around half of the models included emphasis on shared decision-making (56%), mental health (54%), or a guideline/protocol-based approach (46%). We found little direct mention of treatment burden or quality of life (although these are perhaps implied in other categories, such as polypharmacy).

We identified 13 elements for changing *organisation of care delivery*. The majority of models included case management (90%), integration with social and/or community (82%)/secondary care (74%) and a multidisciplinary approach (72%). Around half of the models had elements of home care (54%). Less frequently included organisational changes were co-location of services (33%), scheduled chronic disease appointments (31%), extended appointment times (31%), group visits (21%) or 24/7 support (15%).

We identified eight elements *supporting model delivery*. This most commonly included up-skilling of the primary care workforce (introducing new roles to primary care, 79%), and education of professionals in chronic disease and new ways of working (69%). Around half included some telephone support for patients (49%). Less frequently used elements were funding/payment changes (including incentives or changes to reimbursement mechanisms, 41%), technology (such as clinical IT linkage between services, 38%), risk stratification tools (26%), or tele-health (remote care and monitoring separate from telephone management, 10%).

Change in models of care for multimorbidity over time

Application of our framework showed the following changes over time (see Figure 3).

[insert Figure 3 – Model development over time]

The only statistically significant changes identified were in terms of organisation of care delivery, with a decrease in models implementing home care (-45%), and an increase in models offering extended appointments (+37%). However, there were other trends (≥10% or more change in either direction), tending to favour expansion of primary care services in a single location (e.g. increased co-location of social care services and extended chronic disease appointments), rather than coordination across multiple providers (e.g. decreased care planning and integration with other social/community care services).

Two elements have emerged that did not appear to be present prior to 2010 (trained lay navigator/coaches, and primary care provider networks). The proportion of models utilising each element over time are shown in the Appendix Table A3.

Discussion

Summary

We have presented a framework for reporting and designing models of care for multimorbidity. The framework identifies two foundations, the *theoretical basis*, and *defined target population*; and three categories of *care elements* to implement the model in practice, including (a) clinical focus (b)

organisation of care delivery and (c) support for model delivery. We have provided a list of elements that have been used to date. We have used the framework to identify the focus and gaps in current models, as well as developments over time.

Limitations of the study

The search strategy used to develop the framework was restricted to published models, but was inclusive regardless of study type. However, it is likely that individual elements deployed will continue to change over time, although we expect that the broad framework will endure even if new elements are added. We would highlight that the detail required to develop a usable framework is not the same as that required for replication of models in practice. We would therefore recommend that this framework be considered a minimum rather than maximum level of descriptive detail for model description in future research output (supplementary to existing research reporting checklists).[9] Our framework is not designed to be prescriptive about elements that *should* be used in new models, as the current evidence about 'active ingredients' is so limited. The design and implementation of models will also need to be sensitive to context.[17] However, use of our framework could allow assessment of the contribution of different components in the future, through meta-regression or detailed qualitative process evaluations.[10 18]

Interpretation in the context of the wider literature

The multimorbidity Cochrane review[8] and other published reviews [19] [20] have proposed preliminary classifications of models for multimorbidity, but none have been comprehensive or included new approaches which have not been subject to published trials. Our framework builds on and extends the categorisation used in the recent Cochrane review on effectiveness of multimorbidity models. The National Institute for Health and Care Excellence (NICE) recently issued clinical guidelines for multimorbidity,[21] and our framework complements that by outlining some of the wider changes necessary to support the concept of 'tailored care' in the health system.

Implications of the framework for policy and practice

The theoretical basis of models highlights the assumptions made by the designers. For example, designers may vary in the extent to which they ascribe responsibility for health to individuals or to wider social context. At present, the basis of models appears mostly homogenous, with the most frequent being the CCM. The CCM was initially designed and implemented for single disease-management programmes,[13 22] and there is an argument that this approach is not sensitive to the needs of multimorbidity (e.g. with an emphasis on disease-specific guideline-based decision support).[23] There are fewer models based on more recent developments, such as the 'minimally disruptive medicine' model.[24] Changes in theoretical basis may have significant implications for

model design. For example, models based on 'minimally disruptive medicine' may impact on the clinical focus on self-management, as increased emphasis on self-management may increase the burden of care for a patient.

In terms of population, our mapping of models to the framework identified that the majority of models have focused on a select group, such as older people or those at 'high-risk' (most commonly of hospital admission). The highest-risk patients make up an extremely small proportion of the total population, and of those with multimorbidity, and may already be past the point of care having significant impact on disease course. [25-27] Although older people have more conditions on average, in absolute terms less people with multimorbidity are aged 65 years and over [28] so it is also important that relevant models are developed for younger and lower risk patients. Although these currently less targeted groups do not (yet) account for the majority of secondary care admissions and total cost of care, there may be potential to alleviate their healthcare demand in the future (e.g. through compression of multimorbidity into a smaller period at the end of life). [29] Furthermore, few interventions targeted the most socioeconomically deprived populations, where multimorbidity is known to be more common, occur at an earlier age and is more likely to be related to poor mental health. [28 30]

Our analysis reveals several elements of care which are arguably in need of increased attention. Notably, a mental health focus is lacking from nearly half of all models we included (although this has increased over time). Multimorbid patients with mental health issues are at increased risk for patient safety incidents in primary care,[30] and depression is particularly important in modifying multimorbidity management and outcomes.[31] Few of the models directly focus on treatment burden which is increasingly recognised as an important phenomenon for people with multimorbidity.

Conversely, some of the elements included most commonly do not have a strong evidence base or may be of questionable value in multimorbidity. The emphasis on self-management and prevention may be inappropriate because high levels of morbidity are associated with barriers to self-management[32] and the NICE guidelines found no evidence to support recommendation in multimorbid patients[21]. The potential for prevention in an elderly high-risk population is debatable.[26] Similarly the evidence for case management suggests that patient satisfaction can be improved, but cost and self-assessed health are not significantly affected,[33] particularly for the most complex patients and even when conducted by a multidisciplinary team.[34] Finally, although several models were based on re-organisation of services using tele-health, evidence suggests this may not be a cost-effective approach to managing chronic conditions.[35]

Conclusion

Health systems have only recently begun to implement new models of care for multimorbidity, with limited evidence of success. Careful design, implementation and reporting can assist in the development of the evidence base in this important area. We hope our framework can encourage more standardised reporting and research on the theoretical basis and target population for interventions as well as the contribution of different elements (including interactions between them) needed to provide cost-effective care and support redesign of health systems for those who use them most.[36]

References

- 1. Omran AR. The epidemiologic transition: a theory of the epidemiology of population change. The Milbank Memorial Fund Quarterly 1971;**49**(4):509-38
- 2. Boutayeb A. The double burden of communicable and non-communicable diseases in developing countries. Transactions of The Royal Society of Tropical Medicine and Hygiene 2006;**100**(3):191-99 doi: 10.1016/j.trstmh.2005.07.021[published Online First: Epub Date]|.
- 3. Boyd CM, Fortin M. Future of multimorbidity research: How should understanding of multimorbidity inform health system design. Public Health Rev 2010;**32**(2):451-74
- 4. Nolte E, McKee M. Caring for people with chronic conditions: a health system perspective. 2008. http://www.euro.who.int/ data/assets/pdf_file/0006/96468/E91878.pdf (accessed 01/10/2013).
- World Health Organization. WHO global strategy on people-centred and integrated health services: interim report. 2015.
 http://apps.who.int/iris/bitstream/10665/155002/1/WHO HIS SDS 2015.6 eng.pdf (accessed 01/02/2016).
- 6. NHS England. Five Year Forward View. 2014. https://www.england.nhs.uk/wp-content/uploads/2014/10/5yfv-web.pdf (accessed 01/11/2015).
- 7. Bayliss EA. Simplifying Care for Complex Patients. The Annals of Family Medicine 2012;**10**(1):3-5 doi: 10.1370/afm.1352[published Online First: Epub Date]|.
- 8. Smith SM, Soubhi H, Fortin M, et al. Interventions for improving outcomes in patients with multimorbidity in primary care and community settings. Cochrane Database Syst Rev 2016;4
- 9. Hoffmann TC, Glasziou PP, Boutron I, et al. Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide. Bmj 2014;**348**:g1687 doi: 10.1136/bmj.g1687[published Online First: Epub Date] |.
- 10. Moore GF, Audrey S, Barker M, et al. Process evaluation of complex interventions: Medical Research Council guidance. Bmj 2015;**350**:h1258 doi: 10.1136/bmj.h1258[published Online First: Epub Date]|.
- 11. Davidson P, Halcomb E, Hickman L, et al. Beyond the rhetoric: what do we mean by a 'model of care'? The Australian journal of advanced nursing: a quarterly publication of the Royal Australian Nursing Federation 2006;23(3):47-55
- 12. Arksey H, O'Malley L. Scoping studies: towards a methodological framework. International journal of social research methodology 2005;8(1):19-32
- 13. Bodenheimer T, Wagner EH, Grumbach K. Improving primary care for patients with chronic illness. Jama 2002;**288**(14):1775-79 doi: 10.1001/jama.288.14.1775[published Online First: Epub Date]|.
- 14. Wagner EH. Chronic disease management: what will it take to improve care for chronic illness? Effective clinical practice 1998;1(1):2-4
- 15. Man M-S, Chaplin K, Mann C, et al. Improving the management of multimorbidity in general practice: protocol of a cluster randomised controlled trial (The 3D Study). BMJ Open 2016;6(4):e011261 doi: 10.1136/bmjopen-2016-011261[published Online First: Epub Date]|.
- 16. Levine S, Steinman BA, Attaway K, et al. Home care program for patients at high risk of hospitalization. The American journal of managed care 2012;**18**(8):e269-76
- 17. Craig P, Dieppe P, Macintyre S, et al. Developing and evaluating complex interventions: the new Medical Research Council guidance. BMJ 2008;**337**:a1655 doi: 10.1136/bmj.a1655[published Online First: Epub Date]|.
- 18. Bower P, Gilbody S, Richards D, et al. Collaborative care for depression in primary care. Making sense of a complex intervention: systematic review and meta-regression. Br J Psychiatry 2006;**189**:484-93 doi: 10.1192/bjp.bp.106.023655[published Online First: Epub Date]|.

- 19. Bleich SN, Sherrod C, Chiang A, et al. Systematic Review of Programs Treating High-Need and High-Cost People With Multiple Chronic Diseases or Disabilities in the United States, 2008-2014. Prev Chronic Dis 2015;**12**:E197 doi: 10.5888/pcd12.150275[published Online First: Epub Date]|.
- 20. Bienkowska-Gibbs T, King S, Saunders C, et al. New organisational models of primary care to meet the future needs of the NHS: a brief overview of recent reports. RAND Europe Cambridge 2015
- 21. NICE. Multimorbidity: clinical assessment and management NICE guideline. 2016. https://www.nice.org.uk/guidance/ng56/resources/multimorbidity-clinical-assessment-and-management-1837516654789 (accessed 22/09/2016).
- 22. Bodenheimer T, Wagner EH, Grumbach K. Improving primary care for patients with chronic illness: The chronic care model, part 2. Jama 2002;**288**(15):1909-14 doi: 10.1001/jama.288.15.1909[published Online First: Epub Date] |.
- 23. Boyd CM, Darer J, Boult C, et al. Clinical practice guidelines and quality of care for older patients with multiple comorbid diseases: implications for pay for performance. Jama 2005;**294**(6):716-24 doi: 10.1001/jama.294.6.716[published Online First: Epub Date]|.
- 24. Wallace E, Salisbury C, Guthrie B, et al. Managing patients with multimorbidity in primary care. BMJ 2015;**350** doi: 10.1136/bmj.h176[published Online First: Epub Date]|.
- 25. Wallace E, Smith SM, Fahey T, et al. Reducing emergency admissions through community based interventions. BMJ 2016;**352** doi: 10.1136/bmj.h6817[published Online First: Epub Date] |.
- 26. Roland M, Abel G. Reducing emergency admissions: are we on the right track? BMJ 2012;**345** doi: 10.1136/bmj.e6017[published Online First: Epub Date]|.
- 27. McWilliams JM, Schwartz AL. Focusing on High-Cost Patients The Key to Addressing High Costs? New England Journal of Medicine 2017;**376**(9):807-09 doi: doi:10.1056/NEJMp1612779[published Online First: Epub Date]|.
- 28. Barnett K, Mercer SW, Norbury M, et al. Epidemiology of multimorbidity and implications for health care, research, and medical education: a cross-sectional study. The Lancet 2012;**380**(9836):37-43
- 29. Fries JF. Aging, natural death, and the compression of morbidity. New England Journal of Medicine 1980;**303**(3):130-35 doi: 10.1056/nejm198007173030304[published Online First: Epub Date]|.
- 30. Panagioti M, Stokes J, Esmail A, et al. Multimorbidity and Patient Safety Incidents in Primary Care: A Systematic Review and Meta-Analysis. PLoS ONE 2015;**10**(8):e0135947 doi: 10.1371/journal.pone.0135947[published Online First: Epub Date]|.
- 31. Moussavi S, Chatterji S, Verdes E, et al. Depression, chronic diseases, and decrements in health: results from the World Health Surveys. Lancet 2007;**370**(9590):851-8 doi: 10.1016/s0140-6736(07)61415-9[published Online First: Epub Date]|.
- 32. Bayliss EA, Ellis JL, Steiner JF. Barriers to Self-Management and Quality-of-Life Outcomes in Seniors With Multimorbidities. Annals of family medicine 2007;**5**(5):395-402 doi: 10.1370/afm.722[published Online First: Epub Date]|.
- 33. Stokes J, Panagioti M, Alam R, et al. Effectiveness of Case Management for 'At Risk' Patients in Primary Care: A Systematic Review and Meta-Analysis. PLoS ONE 2015;**10**(7):e0132340 doi: 10.1371/journal.pone.0132340[published Online First: Epub Date]|.
- 34. Stokes J, Kristensen SR, Checkland K, et al. Effectiveness of multidisciplinary team case management: difference-in-differences analysis. BMJ Open 2016;**6**(4):e010468
- 35. Henderson C, Knapp M, Fernández J-L, et al. Cost effectiveness of telehealth for patients with long term conditions (Whole Systems Demonstrator telehealth questionnaire study): nested economic evaluation in a pragmatic, cluster randomised controlled trial. BMJ: British Medical Journal 2013;346 doi: 10.1136/bmj.f1035[published Online First: Epub Date] |.

36. Salisbury C. Multimorbidity: redesigning health care for people who use it. The Lancet 2012;**380**(9836):7-9 doi: 10.1016/S0140-6736(12)60482-6[published Online First: Epub Date]|.

Figure 1: The Foundations Framework

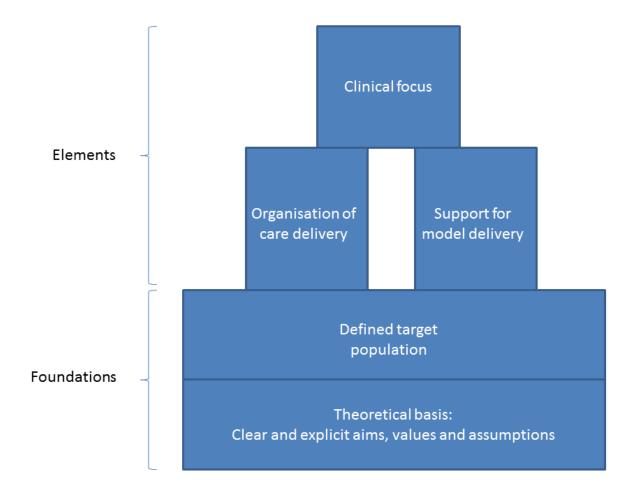


Figure 2: Percentage of models in the current literature using each element of the framework

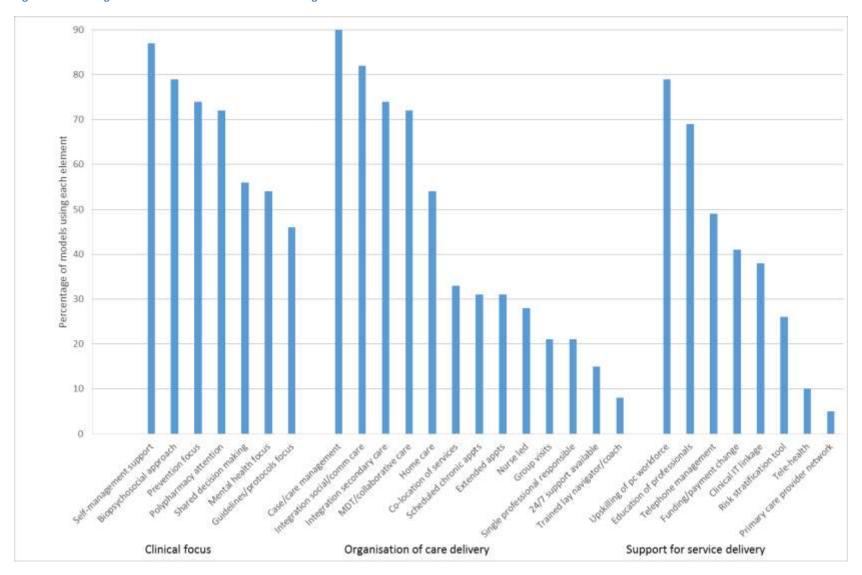
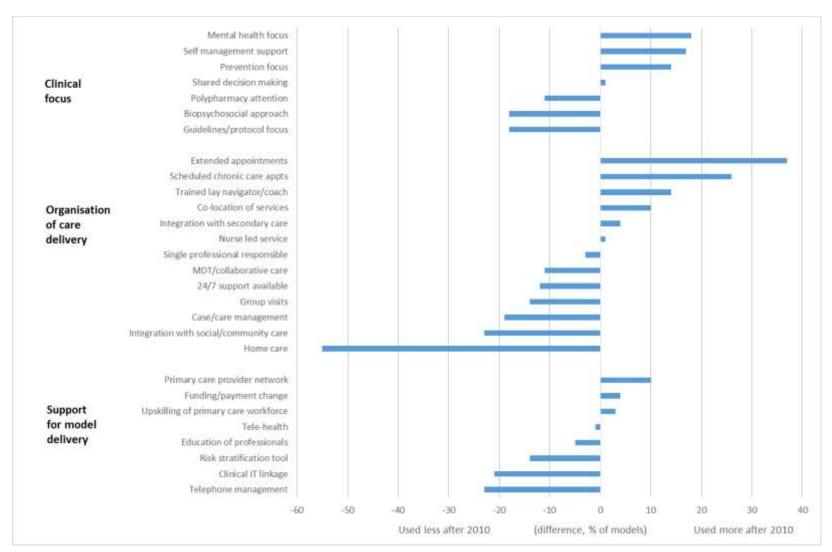


Figure 2: Model development over time. (Difference = % of models described in 2010 or later implementing the element - % of models described prior to 2010 implementing the element)



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Declaration of competing interests

The authors declare no competing interests.

Contributorship

JS, MSM, BG, SM, CS, and PB conceived the study. JS led its development and execution. JS and MSM conducted the initial study screening, selection and extraction. Thereafter, JS, MSM, BG, SM, CS, and PB contributed to various aspects of the empirical work, analysis and presentation. JS, MSM, BG, SM, CS, and PB drafted the manuscript and all authors reviewed it and approved the final version. JS is the guarantor.