Introduction

Multi-morbidity is a major challenge for health and care systems worldwide and of particular relevance for older people. In the UK, two thirds of those aged over 65 have 2 or more long term conditions. [1] In the oldest old (over 85 years of age), living with multiple conditions is inevitable [2] and carries increased risk of adverse consequences for health related quality and duration of life. [3]

The implications of high and increasing prevalence of multi-morbidity challenge researchers, clinicians and policymakers and have stimulated considerable debate. This has emphasised conceptual and practical issues such as, complexity [4,5], evidence gaps, challenges of obtaining robust evidence [4], impact and relationship with specific conditions [3,6,7 ,8] and implications for service delivery. [9] Therefore the mechanisms and consequences of multi-morbidity are key issues for research in an ageing population [4], for older people and their carers.

Although evidence regarding clinical management of multi-morbidity is increasing, considerable uncertainty exists regarding which interventions are most effective [10] particularly in primary and community care settings where most of the care is delivered. [11]

The UK National Institute for Health Research (NIHR) has prioritised the complex health needs associated with multi-morbidity in its calls for funding [12], and associated research topics have been prioritised from a range of national and international perspectives, [4, 13, 14, 15].

Here we report a James Lind Alliance (JLA) Priority Setting Partnership on multiple conditions in later life which identifies the top ten research priorities from the perspectives of older people, their carers, and health and social care professionals.

Method

A multi-stakeholder steering group (n=24) oversaw this Priority Setting Partnership , which followed standard JLA methodology (published in detail elsewhere [16, 17]), modified to meet the needs of older people with multiple conditions.

For a diagram of the process see supplementary materials. Briefly, an initial survey was disseminated to health and social care professionals, carers and older people. The survey results were analysed to identify treatment uncertainties, in the form of researchable questions. A second survey (known as Interim Priority Setting) was then disseminated, to health and social care professionals, carers and older people. This survey detailed the long list of researchable questions and asked participants to list their top ten. The most popular questions were then taken to a final priority setting workshop to decide upon the top 10.

The adaptions to established methodology are described in detail below, and a protocol, [18] including the composition, membership and terms of reference [19] can be found on the project's web pages [20]

Ethical considerations

Although it is important to choose and apply priority setting methods ethically, Priority Setting Partnership s do not normally come under the remit of the NHS Research Ethics Service (NRES), where research priority setting has been seen as service evaluation and development. [21]

Initial Data Collection

The survey was developed and piloted in conjunction with lay members of Voice North (a local public involvement group), many of whom were aged over 80 with multiple conditions. This resulted in changing the wording of the survey. For example, terms such as "multimorbidity" were felt to be incomprehensible by a lay audience and so "multiple conditions" was used. Also, it was suggested that the survey should attempt to portray a positive image of ageing and so the imagery on the survey was changed and phrases such as 'ageing well' were employed.

The purpose of the survey questions was to gather free text responses about treatment uncertainties in respect of symptoms, day to day life, medication and other aspects of treatment and support. A final question asked about questions which did not fit into this framework.

In order to reach a diverse and maximal population of health and social care professionals, older people and carers the survey was advertised widely on-line through social media, academic, professional and charity networks. Participants could complete the survey on-line or request a paper copy. A copy of the survey, formatted for people aged 80+ and their carers, and guidance notes are provided in the supplementary materials; other relevant materials can be found on the project's web site. [20]

Interviews were also conducted to ensure that digitally excluded older people and carers could participate in the Priority Setting Partnership . This face-to-face work was carried out with older

people and carers who either attended community services or were resident in care homes in northeast England. The Interviews were structured to obtain responses to the survey questions and carried out by an experienced qualitative researcher (KL). Potentially eligible participants were identified with the help of community staff with responsibility for their care. They were then approached and the purpose of the proposed interview explained and verbal consent obtained, prior to a conversation structured around the questions asked in the paper survey.   Identifiable information was only gathered when interviewees stated that they would like to be kept informed of the Priority Setting Partnership outcome

Relevant submissions were those which related to the survey questions. Sometimes people did not answer the question in a manner in that could be analysed. For example, when asked about treatment uncertainties some answered with advice about facing the challenges of ageing, rather than questions about their own multiple conditions.

Generating Uncertainties
Data were entered in Microsoft Excel where thematic analysis was performed on all relevant submissions to bring together submissions that had similar meaning. Common issues were grouped into codes. For example, submissions that discussed falling were brought together into the theme 'falls'. The different ways in which falls were discussed were then further analysed, all submissions discussing falls prevention were coded into 'prevention of falls' and all those that discussed fear of falls were organised into 'fear of falling'. These led to questions on falls prevention and a question regarding how best to address fear of falling. Submissions within each code were then further analysed to identify similar issues relating to the primary area of concern, i.e. fall prevention or treatment. These were then grouped together into coded sub-groups. When the researcher (KL) felt that the sub-groups were uniform in context and meaning, sub-groups were synthesised to form researchable questions.

In order to challenge the process of analysis, a data sub-group of the Priority Setting Partnership Steering group including two lay members, discussed emergent codes and the context and meaning of the researchable questions.

Interim Priority Setting
The list of researchable questions was further refined by asking participants (see below) to list (but not rank) the ten questions they thought were the most important.

The Interim Priority Setting exercise was disseminated in the same way as the initial survey. Participation was not limited to individuals that had taken part in the initial survey.

To ensure that digitally excluded older people and carers were also represented within the Priority Setting Partnership face-to-face work was performed with older people and carers attending AGE UK community services in Sunderland who had not participated in the initial survey.   To avoid overwhelming participants with all the questions, the interviews focused on the themes that questions were categorised into. Participants were asked to discuss which of these areas were important to them and why. The researcher linked the participant narrative to the relevant questions and asked the participant if they thought that this was appropriate.

Data from the initial survey, interim priority setting survey and interviews were treated in the same way. The interviews used the same questions as the survey and responses were entered and analysed in the same manner.

The results of the Interim Priority Survey were analysed by vote counting. Each time a question was chosen by a participant it received one vote. To ensure that the differing priorities of health and social care professionals, carers and older people were represented, the top 10 questions from each group were merged into the final list of questions that were taken through to the Final Priority Setting workshop.

Final Workshop
The final Priority Setting Workshop followed standard JLA methodology, conducted over a full day. Drawing on Nominal Group Technique, this involved three rounds of facilitated discussion and prioritisation. The first round involved three small groups comprising a mix of older people, carers and clinicians. The second round was also in three small groups with participants reallocated to create new groups. The final one was a plenary with all participants. While all 21 questions were ranked and re-ranked throughout the day, the end focus was on agreeing the top ten questions that participants agreed were the most important priorities for research.[16,17]. However, it was felt that a full day of discussions may be difficult for older people living with multiple conditions to attend. Therefore, advocates for older people were also invited to attend, defined as individuals who know the issues that older people face every day from personal or professional experience.

Finally, the research priorities of the National Institute for Health and Care Excellence (NICE), and the Academy of Medical Sciences were extracted from published reports [4,9] The priorities of all 3 organisations were categorised and tabulated, to help in the identification of key (shared) priorities and domains.

Results

Initial Data Collection

Data was collected during August and September 2017. In total, 354 people completed the initial survey (answered at least one question). Of these 162 were older people and carers and 192 were health professionals (table 1). Among older people and carers, 15 were interviewed face-to-face.

The analysis of survey responses resulted in 96 researchable questions. These were organised into the following themes: Care provided to older people (n=38); Carers – how to best look after the carers of older people (n=3); Diet & Exercise for older people (n=9); Falls in older people (n=2); Oral health in older people (n=6); Pain in older people (n=1); What the public think of older people (Perceptions of old age) (n=3); How to stay well for longer (Prevention of multiple conditions) (n=1); The emotional and mental health of older people (Psychological wellbeing) (n=4); Sleep (n=1); Loneliness in older people (Social isolation) (n=2), the medicines given to older people (Pharmacy) (n=10) and a miscellaneous category (n=17).

Interim Priority Setting

This took place in March and April 2018 and was completed by 138 participants; 27 older people, 29 carers and 82 health and social care professionals. Of these, 16 older people and 2 carers were interviewed face-to-face. Interviews lasted in the range of 45 minutes to 2 hours.

The amalgamation of the top 10 questions from the differing respondent groups resulted in a list of 21 questions that were then discussed and prioritised in the Final Prioritisation Workshop (see supplementary material).

Final Prioritisation

The final prioritisation workshop was attended by 4 people aged over 80, 5 carers, and 10 health professionals (2 GPs, 2 geriatricians, a Dentist, a Dietician, a Nurse, and Occupational Therapist and a Physiotherapist).

The top ten priorities identified in the workshop are shown in table 2. They included the organisation and delivery of health and social care and the reduction of social isolation as the top priorities for research.

These statements of prioritised research topics are shown also in table 3, alongside the (un-ranked) priorities from NICE and Academy of Medical Sciences , each of which has been classified (broadly) into Organisational, Psycho-Social, Clinical and Biomedical domains.

Discussion

Using the methodologically rigorous JLA process, older people, their carers and health care professionals identified and prioritised key research topics for older people with multiple conditions. Importantly, this exercise highlighted the psychosocial domain, resulting in key priorities about the prevention of social isolation, the promotion of independence and physical and emotional well-being. Clinical priorities of specific importance to older people with long term conditions were also identified, namely exercise therapy, falls (particularly fear of falling), Comprehensive Geriatric Assessment and the recognition and management of frailty.

The findings reported here echo those of several recent research prioritisations in this field. In the UK, NICE and the Academy of Medical Sciences [4, 9] have emphasised optimisation of health and social care systems for the organisation and delivery of essential holistic models of care and prevention and treatment of multiple co-existing conditions. An international consensus exercise with broad stakeholder involvement on research priorities on Ageing in Europe [13], considered multi-morbidity in the context of healthy ageing. Key issues included frailty, organisation and delivery of services, early markers of ill health, disease and function over the life course and effectiveness and efficiency of clinical and social care. A recent American prioritisation for older adults with multiple conditions took the perspective of professional researchers alone and prioritised Health related Quality of Life and its measurement; interactions between medications diseases and outcomes; disability; new models of care; clusters of chronic conditions and the role of caregivers [14]. Many of these research prioritisations focussed on the perspectives of health care professionals. However the JLA process reported in this paper, with its focus on including patients and carers, has additionally highlighted psycho social research priorities important to people living with multi-morbidity and has the person rather than the service as the focus of future research.

A particular strength of the process we used are the adaptions to the standard JLA process to ensure that older people themselves could contribute. This process could be used with potentially hard to reach groups in the future.

Weaknesses of our approach include the issue that for these priorities to be developed further, they will need to be incorporated by research funders in calls for proposals: they are not presented as finalised research questions, and need further development including the most appropriate and preferred research methods and approaches. –Further, while great care was taken to include and ascertain the views of participants in the relevant age group with multiple conditions, we cannot claim that our participants were in other ways representative of the older population in respect of other characteristics (social status and income, ethnicity, education etc)

Conclusion

This paper reports a rigorous JLA process which included older people with multiple conditions as key stakeholders and participants in the identification of research priorities for multi-morbidity in later life. Importantly this process highlighted that psycho-social aspects of living with multi-morbidity, such as prevention of social isolation and promotion of well-being, were key research priorities to older people alongside optimisation of service delivery. It clearly emphasises the benefits of involving older people and their families in setting the research agenda. Future challenges include developing and refining a research agenda that cuts across specialties and agencies and addresses generalist concerns.

2418

Acknowledgements:

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Table 1. **Type of respondent completing the initial survey**

Respondent	Number of responses
People aged 80+	77
Friend / family member of someone aged 80+	68
Other informal carer	2
Former carer	4
Someone aged 80+ that is also a carer	5
Someone that is a 'multiple form of carer'	5
Doctor: specialist in older people's medicine	27
Doctor: GP	13
Nurse	15
Physiotherapist	40
Dentist	11
Dietitian	15
Social Worker	32
Occupational Therapist	9
Pharmacist	9
Doctor: other hospital practitioner	7
Other*	20

* Other includes the following: unknown (n=4); audiologist (n=4); Activity and wellbeing service worker (n=1), care home worker (n=2), deafness charity (n=1), pharmacy technician (n=1); population health and hearing care professional (n=1); retired physiotherapist and carer (n=1); psychotherapist (n=1); rehabilitation officer for hearing (n=1); sheltered / retirement housing worker (n=1); speech and language therapist (n=1).

Table 2. The JLA Priority Setting Partnership top ten priorities for research on multiple long term conditions in later life

1. How can current health, social care and voluntary sectors in the UK be optimised to more effectively meet the needs of older people living with multiple conditions?

2. What are the most effective, cost effective and acceptable ways to reduce social isolation in older people with multiple conditions?

3. What are the most effective, cost effective and acceptable strategies for the prevention of multiple conditions in later life?

4. In what ways can carers of older people with multiple conditions be supported to maintain their own physical and psychological wellbeing?

5. What is the most effective, cost effective and acceptable form of exercise therapy in different health and social care settings with older people with multiple conditions? How does exercise therapy affect outcomes in this population?

6. How can the recognition and management of frailty be improved in older people with multiple conditions? Would this lead to an increase in perceived quality of life?

7. How can Comprehensive Geriatric Assessment be optimally delivered in different patient populations experiencing multiple conditions in older age?

8. What are the most effective, cost effective and acceptable interventions to improve the psychological wellbeing of older people with multiple conditions?

9. How can independent living be most effectively and acceptably enabled in older people with multiple conditions in the UK?

10. How do older people with multiple conditions perceive and manage their risk of falls? How can fear of falling be effectively addressed?

Table 3. Comparison of the research priorities for multi-morbidity identified by NICE, the Academy of Medical Sciences (AMS) and the James Lind AllianceAcademy Priority Setting Partnership on multiple conditions in later life (JLA)*

Organisational	Psycho-Social	Clinical	Biomedical
What is the clinical and cost effectiveness of alternative approaches to organising primary care compared with usual care for people with multimorbidity? (NICE)	What are the most effective, cost effective and acceptable ways to reduce social isolation in older people with multiple conditions? (JLA 2)	What is the clinical and cost effectiveness of stopping preventive medicines in people with multimorbidity who may not benefit from continuing them (NICE)	Is it possible to analyse primary care data to identify characteristics that affect life expectancy and to develop algorithms and prediction tools for patients and healthcare providers to predict reduced life expectancy? (NICE)
What is the clinical and cost effectiveness of a community holistic assessment and intervention for people living with high levels of multimorbidity?(NICE)	In what ways can carers of older people with multiple conditions be supported to maintain their own physical and psychological wellbeing? (JLA 4)	The best ways to prevent the patients developing multimorbidity, and whether this requires different approaches to just preventing individual conditions. (AMS)	The scale and nature of multimorbidity and how it is changing over time. (AMS)
How to organise healthcare systems to deal with multimorbidity more effectively and how best to use digital technology in caring for patients. (AMS)	What are the most effective, cost effective and acceptable interventions to improve the psychological wellbeing of older people with multiple conditions? (JLA 8)	How doctors can increase the benefits and reduce the risks of treatment for patients with multimorbidity. (AMS)	Which clusters of conditions cause the biggest problems for patients. (AMS)
How can current health, social care and voluntary sectors in the UK be optimised to more effectively meet the needs of older people living with multiple conditions? (JLA 1)	How can independent living be most effectively and acceptably enabled in older people with multiple conditions in the UK? (JLA 9)	What are the most effective, cost effective and acceptable strategies for the prevention of multiple conditions in later life? (JLA 3)	The causes of the most common clusters including links with sex, ethnicity, income and lifestyle. (AMS)
How can Comprehensive Geriatric Assessment be optimally delivered in different patient populations experiencing multiple conditions in older age? (JLA 7)		What is the most effective, cost effective and acceptable form of exercise therapy in different health and social care settings with older people with multiple conditions? How does exercise therapy affect outcomes in this population? (JLA 5)	
		How can the recognition and management of frailty be improved in older people with multiple conditions? Would this lead to an increase in perceived quality of life? (JLA 6)	
10 How do older people with multiple conditions perceive and manage their risk of falls? How can fear of falling be effectively addressed? (JLA 10)			

* Source of the recommendations is indicated in brackets (with rank for JLA priorities).

Supplementary material: The 21 questions taken through to the Final Prioritisation Workshop.

1. How can cognitive decline due to lack of sensory stimulation or input be prevented in people who are older and experiencing multiple conditions?

2. How can Comprehensive Geriatric Assessment be optimally delivered in different patient populations experiencing multiple conditions in older age?

3. How can current health, social care and voluntary sectors in the UK be optimised to more effectively meet the needs of older people living with multiple conditions?

4. How can exercises to reduce the prevalence of falls in older people with multiple conditions be effectively incorporated into routine care pathways?

5. How can hospital admission and re-admission rates be reduced for older people with multiple conditions?

6. How can independent living be most effectively and acceptably enabled in older people with multiple conditions in the UK?

7. How can the recognition and management of frailty be improved in older people with multiple conditions? Would this lead to an increase in perceived quality of life?

8. How do older people with multiple conditions perceive and manage their risk of falls? How can fear of falling be effectively addressed?

9. How do older people with multiple conditions perceive their independence? What are the most important factors in maintaining this independence?

10. In what ways can carers of older people with multiple conditions be supported to maintain their own physical and psychological wellbeing?

11. Is there a correlation between poor outcomes in older people with multiple conditions and inadequate levels of care received by them?

12. What are the most effective and acceptable methods to support medication adherence in older people with multiple conditions?

13. What are the most effective and acceptable ways to train and regulate carers of older people with multiple conditions? Would this improve outcomes for this population?

14. What are the most effective, cost effective and acceptable de-prescribing interventions for older people living with multiple conditions?

15. What are the most effective, cost effective and acceptable interventions to improve the psychological wellbeing of older people with multiple conditions?

16. What are the most effective, cost effective and acceptable strategies for the prevention of multiple conditions in later life?

17. What are the most effective, cost effective and acceptable ways to reduce social isolation in older people with multiple conditions?

18. What is the cause and impact of poor sleep on older people with multiple health conditions? What are the most effective ways to address it?

19. What is the impact of social isolation upon the mental and physical wellbeing of older people living with multiple conditions?

20. What is the most effective, cost effective and acceptable form of exercise therapy in different health and social care settings with older people with multiple conditions? How does exercise therapy affect outcomes in this population?

21. What is the prevalence and causes of inadequately controlled pain amongst older people living with multiple conditions?

The Priority Setting Process

Steering group formed

15 specialists in older people's healthcare
4 lay members
1 older person's charity rep
3 data specialists
1 independent chair from the JLA.

359 people from across the UK completed a survey or interview.

236 questions were categorised, merged, summarised & checked against existing research evidence.

97 questions made it to the shortlist and people were asked to pick their top 10

Final workshop completed this process to find the **Top 10** unanswered research questions for Multiple Conditions in Later Life