PEER REVIEW HISTORY

BMJ Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form (http://bmjopen.bmj.com/site/about/resources/checklist.pdf) and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

This paper was submitted to another journal from BMJ but declined for publication following peer review. The authors addressed the reviewers’ comments and submitted the revised paper to BMJ Open. The paper was subsequently accepted for publication at BMJ Open.

(This paper received five reviews from its previous journal but only four reviewers agreed to published their review.)

ARTICLE DETAILS

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<td>AUTHORS</td>
<td>Sidaway-Lee, Kate; Pereira Gray, Denis; White, Eleanor; Thorne, Angus; Evans, Philip</td>
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VERSION 1 – REVIEW

| REVIEWER            | Carl van Walraven  
| University of Ottawa |
| REVIEW RETURNED     | 26-Jul-2017          |

| GENERAL COMMENTS    | I think that this is a nifty study. I tried to do a similar study in the early 2000's but gave up because it was so frustrating (vast heterogeneity between studies, etc). The authors deserve congratulations for their work.
|                     | I have just a few comments about the interval validity of the paper:  
|                     | a) I would provide more detail in the text about the relationship of the time-period that continuity was measured and the time-period in which outcomes were recorded. This is tricky. I think the best way to do this is with continuity measured using time-dependent covariates OR continuity measured up to a specific point after which only outcomes are recorded.  
|                     | b) I would also highlight in the table study design since i believe that cohort studies are much less susceptible to bias than other designs.  
|                     | c) The text (or appendix) should list the patient-level covariates that were adjusted for in the analysis. Sicker patients will have both less continuity and higher risk of death. Therefore, a critical review of how complete the adjustment was for comorbidity is essential. I would consider providing a sensitivity analysis of the study results stratified upon how well patient comorbidity was adjusted for. My hunch is that those with more complete adjustment found less association.  
|                     | d) Page 3, Methods, line 27: sentence starting with 'Any valid...' is unclear to me.  
|                     | I really want there to be a significant association between continuity and decreased risk of mortality. However, I think that we have to be careful with the message of the study. Just because the majority of
studies find an association does not mean that causation exists. It just might mean that all studies are susceptible to the same bias. I believe that all continuity - mortality studies are susceptible to the same strong bias that is discussed in the discussion of the paper, namely that of "reverse causation". Sicker patients are inherently going to have lower continuity for 2 reasons: a) when major health events occur (cancer, cardiovascular disease, trauma, depression), sub-specialists frequently become involved in the patient's care thus decreasing continuity; b) patients whose health needs are not being met by their regular doctor see other physicians to address their unmet health needs. Both of these patients (those with the new health event and those with unmet health needs) are, arguably, more likely to die thereby finding an association between lower continuity and higher risk of death.

Lastly, I believe that continuity of care measures indicate characteristics about the patient that could influence outcomes. Patients who diligently follow the same physician tend to (in my experience) come from higher SES groups and have a greater interest in their health. Such people might have better outcomes because of these characteristics themselves rather than the continuity of care.

Another reason that I disbelieve the causality of the association between continuity of care and decreased mortality risk is the lack of a mechanism. Most people die of cardiovascular disease, cancer, and trauma. It is unclear to me how seeing the same doctor over time is supposed to significantly decrease the risk of any of these death modalities. Is it better BP control? Is it more complete cancer screening? Is it the provision of advice about the use of seatbelts? All maybe but very doubtful. As one who believes that much disease is bad luck and unpredictable, continuity of care stands little chance of changing the risk of these outcomes. This is dour, I know, but (I think) reality. I will change my mind when someone comes out with the properly designed randomized trial to properly determine the association b/w continuity and mortality risk.

REVIEWER
Finlay McAlister
University of Alberta

REVIEW RETURNED
30-Jul-2017

GENERAL COMMENTS
Important question relevant to general readership of BMJ, appropriate study design, conducted as per standard QUORUM criteria, the key studies that I’m aware of in this field were picked up by their literature search, the “what does this study add” box properly summarizes their findings, and the paper is mostly well written (the content in the last 6 paragraphs of the Discussion section could be shortened into 2 focused paragraphs).

One could quibble with the authors’ decision to focus only on studies examining the effect of continuity of care with a physician as opposed to continuity of care with a regular provider, even if that provider is a physician proxy or a multidisciplinary team. In the era of interdisciplinary care I think a sensitivity analysis expanding their eligible studies beyond physician only to physician +/- other team members would be of value (although the results will more than likely be the same).

I would also suggest that they report the results for studies of outpatient continuity of care separately from those studies looking at
inpatient care (ex. weekend vs. weekday attending physician continuity) or studies looking at the transition from inpatient to outpatient care. These are 3 distinct situations.

I think they should consider pooling the outcome data from studies with actual measures of physician continuity (using billing records and summarized with UPC, COC, etc) and not lump them together with satisfaction surveys or patient reports in the Tables. I would agree they can’t meta-analyze all 19 included reports, but that is a subset that they could pool the data from.

I think Table 2 needs to be reworked as the reported results in columns 2-5 weren’t clear and didn’t seem to match the result summaries in the last column for Baker, Bentler, Cerovecki, or Honeyford.

The authors correctly highlight reverse causality as a potential confounder in evaluations of the association between continuity of care and outcomes (since adverse outcomes may disrupt continuity of care due to the need for involvement of disease-specific specialists to deal with the adverse outcomes). However, another important source of bias in observational studies examining continuity of care is time dependent bias and I think the authors should discuss which of the studies they included took measures to mitigate this risk (such as the use of time varying covariate analyses, landmark time analyses, etc). See van Walraven et al in J Clin Epidemiol. 2004 Jul;57(7):672-82 and Austin et al in J Eval Clin Pract. 2006 Dec;12(6):601-12 for a fuller description of the issue and potential analytic approaches.

Rather than just listing the different continuity measures used, it would be helpful to include a few sentences on the advantages vs. shortfalls of the more common methods used in the literature (UPC, COC, etc).

In the discussion section they mention that they think publication bias is unlikely as “there are less likely to be financial incentives for suppressing studies with an unfavourable conclusion”. However, financial incentives are not the only cause of publication bias – authors and journal reviewers/editors are less enthusiastic about negative studies even with orphan drugs/interventions and in a world of limited time the natural human reaction is to write up your positive studies (or interesting negative ones) first.

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**Reviewer**

Chris van Weel  
(1) Radboud University, Nijmegen, The Netherlands; (2) Australian National University, Canberra, Australia

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**General Comments**

Comments to authors  
This paper is a systematic review of studies on continuity of care and mortality. As premature mortality can be considered the key indicator of outcome of care, this study addresses a key feature of continuity. Given the fact that the authors did not find any systematic review in their search of the literature, this is in all probability the first review assessing the relation continuity – mortality in its kind. That makes it a very important paper. The authors have done a robust search of the literature, and analysed their data in a careful way, assessing the quality and reliability of the data they have found. The heterogeneity of the
studies they found, made it impossible to pool data, and as a consequence the findings are presented in a narrative way, accounting for the variation in studies. On average, the primary studies are of a good quality, which adds to the relevance of this paper. The authors present and discuss their findings in a reflective way, avoiding cause-consequence conclusions from the associations between continuity and mortality. Only based on a broader range of arguments do they tentatively present their study as supporting the protective effect of continuity of care for patients’ health.

I have three points I would like the authors to look into:

1. My first point is the variation on observation time between the studies. It is difficult to accept that continuity of care could have an influence of individuals’ health status, when it was only adhered to during a short period of time – at least there is no conceivable argument to warrant a ‘short-term effect’. And it looks to me that particularly situations where patients were moved due to their deteriorating health status (end of life care in hospice; nursing home care for frail elderly) would present conditions for ‘short term’ studies on continuity. In these settings, continuity of care is of eminent importance, but it can not be expected to contribute to or be related to enhanced survival. In other words, studies with a short observation span might be liable to an inverse relation between continuity of care and survival. From this perspective it would be interesting to compare the studies with the longer and the shorter observation time in terms of their association: would the findings have been different if a minimum observation time had been applied and only the long term studies had been included in the analysis?

2. The studies come from a variety of settings, in which the health system has arranged for continuity of care. In the UK and the Netherlands (and probably Israel and Croatia) the ‘primary care structure’ and the role of listing to a general practice has provided a structure that promotes continuity of care. This is quite different from the US, where this very much depends on the insurer involved. It can be expected, therefore, that the room for variation in studies from the US (and probably Canada) is much larger than in studies from the UK, the Netherlands. And consequently show a stronger association with mortality. Would it be possible to use the ‘health structure setting’ as a determinant of the correlation?

3. The study is based on analysis of primary studies that included physicians and I understand that four studies were found but excluded because nurses were the main provider of care, or a team. I understand why the authors excluded these studies, but on the other hand it is a pity to exclude their information. I suggest that the authors provide a bit more information:
   a. In care through a team, one would assume that a physician was part of the team, and why not include this in the 19 other studies?
   b. What was the direction of the association continuity-mortality in the nurse-led care? It might be possible – following the arguments the authors provide to substantiate their decision to exclude these studies – that ‘medical decisions’ that influence life expectancy had a less prominent place and consequently the correlation was less distinct. But it would be at least remarkable if nurse-continuity would lead to an inverse relation. That is, unless the nurse interventions were directed at care situations of patients with limited life expectancy. It would be an enrichment of the paper if information of nurse-led care was included in the discussion.
Comments to the editor
This is an important study, that provides empirical data on the direction of health reforms to improve the outcome of health care. For that reason I strongly recommend publication of this paper. The paper supports the policy to strengthen primary health care as the core component of the health system. And it provides valuable information of how to measure and monitor health system reform – the current discussion on monitors of primary health care.
At the same time, the paper stresses the importance of continuity of (sub)specialist care as well. This is an ignored aspect and the paper invites further thought on this. It stresses in my view the importance of primary care – specialist care coordination and extending the involvement of specialists in the continuity loop. For that reason I think this paper would deserve a commentary to accompany its publication, addressing these points.

REVIEWER
Christine Morgan
NHS Stockport CCG & Coalition 4 Collaborative Care

REVIEW RETURNED
11-Aug-2017

GENERAL COMMENTS

• Are the study's aims and the issue and questions that the paper addresses relevant and important to you as a patient? Do you think it would be relevant to other patients like you? What about carers?

I think the objective in looking at '...whether there is a relationship between the receipt of continuity of doctor care and mortality.' is relevant, important and timely to me as a patient and to carers too. In England and most of the globe healthcare systems are striving to provide value for money and keep up with technological and medical advances. Equally with rising demand new and different roles are being brought into healthcare in order to deflect time away from overworked doctors as well as ‘scaling up’ care this study brings into question whether there is a danger that these approaches may well threaten continuity of doctor care and conversely worsen patient outcomes through decreasing the interpersonal component of care.

The paper acknowledges that much is known about how continuity of care in general practice has positive outcomes which include patient satisfaction, adherence to medication and reductions hospital use but before this study there has been no attempt to see if there was any correlation between continuity of any doctor with mortality outcomes.

• Are there any areas that you find relevant as a patient or carer that are missing or should be highlighted?

I think the fact that this study looks at ‘any doctor’ i.e. not just those in General Practice and ‘any patient group’ ‘...in any setting’ is significant and could be highlighted although it is clear in the text. I don’t think there are any areas that are missing but I think that subsequent studies could usefully segment patient populations to get down to more granular levels of details and I would welcome seeing the figures and data relating to patients with Long Term Conditions to see if enhanced continuity of care correlates with quality of life as well as decreased mortality.

The fact that continuity of care from ‘any’ doctor in ‘any’ setting is also significant as perceived wisdom in England appears to be that only continuity of care by GPs is significant and that GPs hold the
ring of continuity even when patients are treated by other patients in other settings which I've never been personally convinced by in practice.

• From your perspective as a patient, would the treatment, intervention studied, or guidance given actually work in practice? Is it feasible? What challenges might patients face that should be considered?

As this is a systematic review of existing studies there is no treatment or intervention as such in this study.

• Are the outcomes that are being measured in the study or described in the paper the same as the outcomes that are important to you as a patient? Are there others that should have been considered?

I think that mortality rates measured against continuity of care of doctor is a very pertinent outcome and as described as ‘the most important and serious of all outcomes.’ I also think that it would be useful to know if doctor satisfaction correlates with patient satisfaction, experience and mortality rates too? For me personally as someone with Long term Conditions I am also interested in personal and social quality of life outcomes, prior to death, and how the interpersonal continuity of care by doctors has an impact on that as perceived by the patient against agreed outcome measures. Patient experience defined as including safety and quality of clinical and social interventions is often seen as of a lesser value than pure clinical outcomes delivered by any doctor or clinician and I’m pleased to see how this study links continuity of care by doctors as a key component with mortality rates. This then makes me hypothesise that the basic human effect of continuity of care has linked clinical outcomes too.

• Do you have any suggestions that might help the author(s) strengthen their paper to make it more useful for doctors to share and discuss with patients?

I think this is a very important study which underlines the importance of the basic human effect of continuity of care of doctors and paves the way to further studies of other health care practitioners where continuity of care is essential to good patient experience and good outcomes such as midwives for example.

This study could be of immense value to doctors to share with their patients as a tool to consider alongside the introduction of for example New Care Models, in England, where planned effectiveness could compete with continuity of care from a doctor and might potentially adversely affect patient outcomes. The fact that this study was the idea of a patient, working in partnership with researchers, underpins its value as something that really matters to patients and is the first study I have seen of its kind.

VERSION 1 – AUTHOR RESPONSE

Reviewers’ Comments to Author:

Reviewer: 1
Reviewer Name: Christine Morgan  
Institution and Country: Acting as Independent patient reviewer in the UK  
Competing Interests: Independent current roles include: Non Executive/Lay member of NHS Stockport CCG - remunerated role, Co-production team member of the Coalition for Collaborative Care - Expert Advisor involvement fee paid for sessions

Please see below review as copied in Word (PDF file of review also attached)


• Are the study's aims and the issue and questions that the paper addresses relevant and important to you as a patient? Do you think it would be relevant to other patients like you? What about carers?

I think the objective in looking at '..whether there is a relationship between the receipt of continuity of doctor care and mortality.' is relevant, important and timely to me as a patient, other patients and to carers too. In England and most of the world healthcare systems are striving to provide value for money and keep up with technological and medical advances. Equally with rising demand new and different roles are being brought into healthcare in order to deflect time away from overworked doctors as well as 'scaling up' care this study brings into question whether there is a danger that these approaches may well threaten continuity of doctor care and conversely worsen patient outcomes through decreasing the interpersonal/human component of care.

The paper acknowledges that much is known about how continuity of care in general practice has positive outcomes which include patient satisfaction, adherence to medication and reductions hospital use but before this study there has been no attempt to see if there was any correlation between continuity of any doctor with mortality outcomes.

• Are there any areas that you find relevant as a patient or carer that are missing or should be highlighted?

I think the fact that this study looks at 'any doctor' i.e. not just those in General Practice and 'any patient group' '..in any setting' is significant and could be highlighted although it is clear in the text. I don't think there are any areas that are missing but I think that subsequent studies could usefully segment patient populations to get down to more granular levels of detail which is partly demonstrated in Table 1 where information re specific conditions is detailed. I would welcome hearing about a patients’ reported outcomes of their care and their perceived personal and social outcomes when this care has continuity and when it has not.

The fact that continuity of care from 'any' doctor in 'any' setting is also significant as perceived wisdom in England appears to be that only continuity of care by GPs is significant and that GPs hold the ring of continuity even when other doctors in other settings treat patients, which I've never been personally convinced by in practice.

• From your perspective as a patient, would the treatment, intervention studied, or guidance given actually work in practice? Is it feasible? What challenges might patients face that should be considered?

As this is a systematic review of existing studies there is no treatment or intervention as such in this study.
Are the outcomes that are being measured in the study or described in the paper the same as the outcomes that are important to you as a patient? Are there others that should have been considered?

I think that mortality rates measured against continuity of care of doctor is a very pertinent outcome and as described as ‘..the most important and serious of all outcomes.’ I also think that it would be useful to know if doctor satisfaction correlates with patient satisfaction, experience and mortality rates too?

For me personally as someone with Long term Conditions I am also interested in personal and social quality of life outcomes, prior to death, and how the interpersonal continuity of care by doctors has an impact on that as perceived by the patient against agreed outcome measures. Patient experience defined as including safety and quality of clinical and social interventions is often seen as of a lesser value than pure clinical outcomes delivered by any doctor or clinician and I’m pleased to see how this study links continuity of care by doctors as a key component with mortality rates. This then makes me hypothesize that the basic human effect of continuity of care has linked clinical outcomes too, described later in the study as ‘..an optimism boost for health.’

Do you have any suggestions that might help the author(s) strengthen their paper to make it more useful for doctors to share and discuss with patients?

I think this is a very important study, which underlines the importance of the ‘basic human effect’ of continuity of care of doctors and could pave the way to further studies of other health care practitioners where continuity of care is essential to good patient experience and good outcomes.

This study could be of immense value to doctors to share with their patients as a tool to consider alongside the introduction of for example New Care Models, in England, where planned efficiencies could compete with continuity of care from a doctor and might potentially adversely affect patient outcomes.

In the concluding section on possible further implications the case is well made that where continuity of care of a doctor with their patient occurs there is evidence of:

1. Increased patient satisfaction
2. Increased take-up of health promotion
3. Reduction in use of hospitals

The above 3 potential implications are important for doctors to realise on this influence both with their patients and to influence policy makers to ensure that ‘the human side of medicine’ is not neglected in quality of health and ultimately effects on mortality.

We are glad the patient representative is so positive about the article.

Reviewer: 2
Reviewer Name: Carl van Walraven
Institution and Country: Ottawa Hospital Research Institute, Canada
Competing Interests: None

The authors do a great job with a very difficult task (identifying all studies examining the association of physician continuity on patient survival).

MAJOR
The major limitation of these studies is their observational design. This results in the distinct possibility of reverse causality in which the process which eventually kills a person actual causes discontinuity. In such cases, analyses will conclude that discontinuity is associated with an increased death risk. Neither baseline covariate adjustment nor time-dependent covariable measures of continuity can address this issue. Therefore, in the absence of a randomized trial (which will likely never be done), I believe that any statement implying that physician continuity of care causes increased survival cannot be justified. I think that publishing this systematic review without prominently highlighting this major issue regarding these studies would be misleading.

We had identified Professor van Walraven as a leading thinker on our subject before we originally submitted. He has written about the possibility of reverse causality and he repeats this concern in his assessment. We already included sentences stating clearly that all these data are observational and so the possibility of reverse causality and time dependent bias needs consideration. We have now also added extra sentences on this to the introduction. We have also set out how much work the authors of many of the articles which we have reviewed have done to counter this theoretical problem and we now state that 63% have referred to it and worked to mimitise it.

Reviewer: 3
Reviewer Name: Chris van Weel
Institution and Country: Emeritus Professor of Family Medicine/General Practice, Radboud University Nijmegen, The Netherlands, Honorary Professor of Primary Health Care Research, Australian National University, Canberra, Past President of World Organization of Family Doctors WONCA
Competing Interests: None

The is an excellent and much needed paper, looking into the impact of continuity of care on individuals’ health status. General practitioners and other primary health care professionals consider continuity of care as their professional hallmark [1] and patients value it. But for too long, health policy has approached continuity of care as an ornament rather than an instrumental feature of the system in fostering population health. This review shows that it is time to change this and secure continuity of care and long-standing relations between professionals and patients at the core of contemporary health systems.

The authors have approached their findings critically, with due consideration of bias. As they state, no RCTs were found and theoretically the RCT model would provide the most convincing evidence. Against this one might argue that few aspects of health policy can boast the sophistication of RCT-generated evidence. And other available research in the absence of RCTs should better be embraced – as the authors have done.

The paper raises at least two important questions: the first is in what way continuity and the personal relation over time contribute to individual health. In this context it might help to consider the ‘paradox of primary care’: the finding that when measured with disease-specific indicators, specialists may be superior to generalists, but when individual health status is used as outcome, generalists do better [2]. From this paper can be concluded that continuity of care is a core component of this paradox that will bring a relation of trust but also health education, prevention, early diagnosis, and integration, coordination and prioritisation of care between various episodes of illness, into play in caring for patients. This stresses the need to better understand their mechanisms – how they contribute to health – to further improve care of patients.

The leads to the second point: when continuity of care – and the other above factors – are a contributing factor to health, it must be assumed that the quality matters and there are good, positive, as well as poor, disruptive ways of performance. Better understanding of what makes continuity of care the contributing factor for effective care of patients is a priority for the professional development, teaching and training of current and future (primary) health care professionals.

1. WONCA Europe. The European definition of general practice/family medicine. Wonca Europe
Professor van Weel has an international reputation and has recently been the President of the World Association of Family Doctors. We are therefore particularly pleased he is so enthusiastic about our article.

He suggests we might cite Stange et al. (2009) “Paradox of Primary Care”. We agree this is a classic reference in family practice/primary care.

However, it emphasizes the difference between doctors in primary care and secondary care, whereas our new finding is that continuity provided by doctors both in primary care/general practice and in secondary care is associated with reduced mortality for patients. This is therefore a unifying not a contrasting finding theme. Therefore, we have not included this reference.

Track changes

We included a version with the changes made tracked with our resubmission so you can see them clearly.

**VERSION 2 – REVIEW**

<table>
<thead>
<tr>
<th>REVIEWER</th>
<th>Christine Morgan</th>
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<tbody>
<tr>
<td>Acting as Independent patient reviewer in the UK</td>
<td>10-Jan-2018</td>
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**GENERAL COMMENTS**

Please see below review as copied in Word (PDF file of review also attached)


- Are the study’s aims and the issue and questions that the paper addresses relevant and important to you as a patient? Do you think it would be relevant to other patients like you? What about carers?

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REVIEWER
Carl van Walraven
Ottawa Hospital Research Institute

REVIEW RETURNED 16-Jan-2018

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The authors do a great job with a very difficult task (identifying all studies examining the association of physician continuity on patient survival).

MAJOR
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REVIEWER
Chris van Weel
Professor Chris van Weel
Emeritus Professor of Family Medicine/General Practice
Radboud University Nijmegen, The Netherlands
Honorary Professor of Primary Health Care Research,
Australian National University, Canberra
Past President of World Organization of Family Doctors WONCA

REVIEW RETURNED 18-Jan-2018

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of continuity of care on individuals’ health status. General practitioners and other primary health care professionals consider continuity of care as their professional hallmark [1] and patients value it. But for too long, health policy has approached continuity of care as an ornament rather than an instrumental feature of the system in fostering population health. This review shows that it is time to change this and secure continuity of care and long-standing relations between professionals and patients at the core of contemporary health systems.

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The leads to the second point: when continuity of care – and the other above factors – are a contributing factor to health, it must be assumed that the quality matters and there are good, positive, as well as poor, disruptive ways of performance. Better understanding of what makes continuity of care the contributing factor for effective care of patients is a priority for the professional development, teaching and training of current and future (primary) health care professionals.


authors of many of the articles which we have reviewed have done to counter this theoretical problem and we now state that 63% have referred to it and worked to minimise it.

Third assessor Professor van Weel

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He suggests we might cite Stange et al (2009) “Paradox of Primary Care”. We agree this is a classic reference in family practice/primary care. However, it emphasises the difference between doctors in primary care and secondary care, whereas our new finding is that continuity provided by doctors both in primary care/general practice and in secondary care is associated with reduced mortality for patients. This is therefore a unifying not a contrasting finding theme. Therefore, we have not included this reference.

Track changes
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