Author’s response to reviews

Title: Exploring the usefulness of Lexis diagrams for quality improvement

Authors:

Sara Dahlin (sara.dahlin@chalmers.se)

Version: 1 Date: 17 Nov 2019

Author’s response to reviews:

Dear Editor,

Below is my response to the reviewers' comments, including extracts from the updated manuscript with reference to the line numbers, when possible.

Yours sincerely,

The author

Reviewer reports:

Julie Reed, Dr. (Reviewer 1): Thank you for the opportunity to review this interesting manuscript that I think would be of interest to specialists in this field. I think this paper could be suitable for publication but requires major revisions.

I am not an expert in survival probability or lexis diagrams, but am very interested in the practical use of data (and the need to appropriately visualize complex information) to drive action and have conducted extensive qualitative research on the relationship between data use and actions taken.

I think that the issue of data visualization and interpretation is very important to the field of quality improvement. As such I was very interested to read this paper and to learn of a new method.

However, I found at times the text to be hard to follow as it assumed a lot of prior knowledge and failed to appropriately explain key concepts so that they can be easily followed. This was particularly an issue in the introduction, and in general I felt I had to do a lot of work to make sense of the manuscript (not helped by having to scroll up and down to the figures which I appreciate is out of the authors control).
I have put detailed comments below about where I struggled to understand or make sense of the manuscript with suggestions for how this can be improved. I don't believe this is necessary a flaw of the study or interpretation, but more the clarity required to present new complex ideas in a way that make sense to others in the field.

Author response: I highly appreciate the time you spent on detailed reviewing and all helpful comments. Thank you very much!

I did have one major concern regarding the study validity. Methodologically the paper would be stronger if others who hadn't been involved in constructing the diagrams were then exposed to the diagrams with either verbal and/or written information to see their reactions and ability to interpret. The diagrams are not straight forward and I wonder how frontline staff would have responded to them. Clearly the group who developed them have spend significant time and investment therefore biased in perceived value. Indeed in one of the quotes an interviewee mentions that they perceive that other staff would value seeing the data (how long patients live for) - this could be tested empirically which would greatly strengthen the paper.

Author response: Following text has been added to the limitations section of the discussion (line 483-491): ‘Although the researcher constructed all visualizations while the process owners contributed with domain knowledge, the iterative analysis increased the method understanding among process owners over time. Therefore, the process owners may have been somewhat biased in their interpretation of the method’s usefulness towards the end as compared to future novel users, and it should be noted that education and experience may be needed to fully grasp the method, preferably using an interactive version of the Lexis diagram. Future research on Lexis diagram usefulness is encouraged also to present Lexis diagrams to practitioners not included in the construction of the diagrams and gain insights about their perceptions of Lexis diagram usefulness.’

Further empirical data collection is unfortunately not possible within this study.

Detailed comments:

Abstract

Methods: suggest for clarity change sentence order -

Lexis diagrams were produced based on data from…

The usefulness of Lexis Diagrams was explored through…

Iterative development of the diagrams…

Author response: The method section of the abstract has been updated (line 32-37): ‘Lexis diagrams were produced based on data from a gynecological cancer quality registry (4481
patients). The usefulness of Lexis diagrams was explored through iterative data identification and analysis through semi-structured dialogues between the researcher and domain experts (clinically active care process owners) during five meetings. Visualizations were produced and adapted by the researcher between meetings, based on the dialogues, to ensure clinical relevance, resulting in three relevant types of visualizations.’

Results:

First line belongs in methods.

I found the results confusing - not having prior knowledge of what Lexis diagrams do, nor having read the paper. This needs work to make the findings clearer to someone who is not an expert in this approach. e.g. stating the basic principles of what a lexis diagram shows and the extent to which users found this helpful.

Author response: The Lexis diagram has been clarified in the Background section of the abstract line 25-29): ‘However, there is a lack of QI methods visualizing multiple, related factors such as diagnosis date, death date, and cause of death to unravel their complexity, which is necessary to understand processes related to survival data. Lexis diagrams visualize individual patient processes as lines and mark additional factors such as key events.’

The extent to which users found this helpful has been clarified in the conclusions section of the abstract (line 44-46): ‘Lexis diagrams can aid understanding of survival data, triggering important dialogues between care givers and supporting care quality improvement and new perspectives, and can therefore complement survival curves in quality improvement.’

Introduction:

I found the text from line 59 - 67 introduced intriguing in concepts, but it was hard to understand the logic connecting the statements and the meaning the author would like the reader to take from them. I think all the points raised are relevant but a little more "hand holding" to explain the meaning of the points in relation to the authors overall argument, perhaps with an example, would help make this section clearer.

e.g. Why does system complexity and non-linearity matter when building knowledge to guide local QI efforts - and how specifically does this relate to the use and presentation of data?

I feel I am trying to imagine/guess the connections and direction of the authors argument.

Author response: The background has undergone major revisions, based partly on your review comments. The text including above-mentioned concepts now goes (line 93-105): ‘However, survival data documented in health information systems typically include several important factors, such as time of diagnosis, time of death and cause of death, which may need to be understood simultaneously, as they together reflect care process complexity. Notably, the level
of complexity in a (care) system depends on the number of components and their interrelatedness (19), and non-linearity also characterizes complex system behavior (20); studying relevant interrelatedness between components within the care system may support understanding of the system (19) and thus aid local improvement action (2, 3). Characterizing components’ interrelatedness becomes necessary to unravel the system’s complexity (19), and domain experts may in this way be supported in their understanding of local care process complexity and thereby guided into QI efforts (21). Such data, however, do not fit into a control chart visualization, and therefore another method is needed, where focus instead lies on understanding and visualizing the complexity of several process-related factors for feedback purposes.

What does "complexity harnessed in data documentation" mean? Can you explain or provide an example?

Author response: This text is revised to: “However, survival data documented in health information systems typically include several important factors, such as time of diagnosis, time of death and cause of death, which may be need to be understood simultaneously, as they together reflect care process complexity.” (line 93-95)

Why do control charts not represent complexity? Some experts would argue that they do - so you need to be clearer in your reasons for this statement.

Author response: This has been revised, the reasoning around why not to use control charts is: ‘However, survival data documented in health information systems typically include several important factors, such as time of diagnosis, time of death and cause of death, which may need to be understood simultaneously, as they together reflect care process complexity. … Such data, however, do not fit into a control chart visualization, and therefore another method is needed, where focus instead lies on understanding and visualizing the complexity of several process-related factors for feedback purposes.’ (line 93-95, 102-105).

I'm not sure control charts assume that people know the related processes - QI would often advocate for other methods to better understand processes that should be used in conjunction with control charts, of which searching medical records would only be one.

Author response: The reasoning around using additional methods has been deleted.

Line 73 - I feel this sections jumps to two new topics - statistical significance and survival data.

I think it would be helpful to introduce this section more clearly - e.g. One area in which improvements could be made in the visual representation of data is patient survival probability… Explain what patient survival probability is and why important (assuming no prior knowledge of the reader), Explain how data normally represented and why the data could be better represented…
It would be helpful if the author could specify information about the other visualisation methods mentioned (given this is the key point of the article) rather than assume the reader knows or requiring the reader to search for the information. e.g. what are the key features of Kaplan-Meier curves in visualising survival probability? What are the strengths and limitations of such methods in visualising data for what purposes (prediction, action etc)? Could an example K-M diagram be included alongside the sample Lexis diagram?

Author response: Due to the major revision, the argumentation has been changed. Further, the visualization methods are now presented in a hopefully clearer way throughout the background section and through the example visualizations of all three types of methods in Figure 1, as well as through Table 1, which is in line with your next comment (please see Background, Figure 1 and Table 1).

Could a table be presented in which the key features and benefits/limitations of the two diagrams could be compared side by side? and perhaps include control charts as a third visualisation method given its dominance in QI and the authors dismissal of this method.

Author response: A table comparing the three methods is included (see Table 1).

Figure 1 - it would be helpful for the author to describe a couple of the lines in detail so the reader has a clear sense of how to interpret the lines and what information can be gleaned from the diagrams. This could be done in main text or in as Fig text.

Author response: The figure text is changed to support interpretation: ‘Figure 1. Examples of different visualizations. 1a. Survival curve comparing two cohorts, from Dahm-Kähler et al. (15). 1b. Control chart, with upper and lower (statistical) control limits. 1c. Lexis diagram, presenting data of four hypothetical patients, each patient represented by a lifeline starting on diagnosis date (x=0) and continuing upwards until patient’s death, or extending to right border if patient is alive at data extraction. The lifelines’ 45-degree angle reflects equal time passing along both time axes in the Lexis diagram. Dots represent time of death, and crosses show other events occurring between diagnosis and death.’

Line 105 - “this versatility enhances understanding” - this needs a lot of unpacking - it is not immediately clear to me why this is. It would be helpful to understand more about the types of trends and patterns that are easy to identify using these diagrams and how this is done.

Author response: I have tried to clarify in the background, and also explain in the method (see your comment regarding Line 233 in the previous draft). On line 116-128): ‘Lexis diagrams are one such as possible method, used to identify joint effects of age, period, and cohorts (24, 25), and serve as a possible complement to subsequent statistical survival analysis (26) In its basic configuration, a Lexis diagram visualizes individual lifelines along two time axes: calendar time and age or year since diagnosis (27-29), where the lines extends in a 45-degree angle as time passes along both x and y axes. Figure 1c shows a basic Lexis diagram with hypothetical
patients. Lines and markings can be differently colored or shaped to represent different attributes (30, 31), for instance, line colors representing diagnoses; marking shapes representing care process events, and marking colors representing event attributes. New data (lines/patients or markings) can be continuously added, supporting timeliness in analysis. Lexis diagrams can for example be analyzed by counting markings, such as dots (30) or lines, across a section of the diagram (32), to identify patterns. Together, this versatile visualization may reveal the complex pattern of events, time between events and attribute data, that may enhance understanding (33).

Line 107 - it would be helpful to explain more clearly how the Lexis diagrams can help with prediction, timeliness and complexity. Not immediately clear.

Author response: The text has been revised and prediction has been changed to feedback by following data over time: Line 120-128):” Figure 1c shows a basic Lexis diagram with hypothetical patients. Lines and markings can be differently colored or shaped to represent different attributes (30, 31), for instance, line colors representing diagnoses; marking shapes representing care process events, and marking colors representing event attributes. New data (lines/patients or markings) can be continuously added, supporting timeliness in analysis. Lexis diagrams can for example be analyzed by counting markings, such as dots (30) or lines, across a section of the diagram (32), to identify patterns. Together, this versatile visualization may reveal the complex pattern of events, time between events and attribute data, that may enhance understanding (33).

Line 133-137: ‘(34). With potential to elicit feedback by (e.g.) presenting trends in data (35), ensure timeliness through real-time monitoring (32), and visualize complexity through the use of different colors, markings, and time axes (33), they may help practitioners understand care given and improve actionable survival data analysis.’

Line 141 - I am still not clear why timeliness is an issue for traditional analysis, nor how this issue is mitigated using lexis diagrams - can this be explained? I assume relates to the time taken to collect sufficient sample size and then conduct analysis as a one-off event? Whatever the reason this needs to be much clearer.

Author response: The revised text is:

Regarding traditional analysis Line 69-82: ‘Such data are often analyzed through survival curves and used for evaluating improvement efforts (see e.g. 14), but survival curves have another purpose; they contribute to “global knowledge” useful in (e.g.) medical research such as clinical trials of new treatments, or, as in Dahm-Kähler et al. (15), showing that centralization of surgery improves survival on a population level. Kaplan–Meier analysis is the most popular type of survival data analysis (16), resulting in a survival curve plot (exemplified in Figure 1a) featuring time since an event on the x-axis and a survival probability measure on the y-axis, such as survival rate (15) or cumulative survival (16), presenting the estimated survival rate of (e.g.) a patient group with a particular diagnosis over a certain time interval since the starting event. For further information about Kaplan–Meier curves, see Jager et al. (16). Its purpose is thus not to
support local process understanding and action for QI and due to the need for large sample sizes and aggregation of data, other methods are needed to ensure timeliness and gaining feedback by following data over time, contributing to local improvements.’

Regarding Lexis diagrams and timeliness (Line 124-125): ‘New data (lines/patients or markings) can be continuously added, supporting timeliness in analysis.’

Line 142 - it is not clear how Lexis diagrams relate to before/after analysis -so the "third" reason is confusing - can this be explained earlier.

Author response: The text about before and after analysis is deleted, including the “third” reason for choosing gynecological cancer data.

Line 162 - I am not familiar with the term "process owner" - can you explain what this means?

Author response: The revised text (line 160-164): Designated process owners, who are clinically active physicians with great practical knowledge, are “owning” the process by being responsible for developing the care pathway for a specific cancer diagnosis, making them invaluable for quality improvement collaborations involving method assessment and development.

Line 223 - its not clear to me why the line representing surviving patients goes down and backwards (right to left) - this implies going backwards in time? I would have thought that the line would go down and forwards (left to right)? Useful to explain this convention.

Author response: The lines went backwards due to a programming issue. However, the figure has been deleted due to this problem, the comment nr 4 by reviewer 2 and a shift towards less comparison between methods.

Line 233 - in this paragraph are you essentially say that you count the number of dots between certain lines? If so is it helpful to quantify the number of dots for the cells you are talking about to help illustrate the point? Would this be standard practice? Or are you just meant to eyeball the graph?

Are these figures statistically significant? If not how are you comparing the results to the statistical survival curve? This also suggests that there is a difference in survival rates from the beginning of the timeline?

These claims need more precision.

Author response: There are different ways to identify patterns, which is clarified as follows:

On line 125-127: ‘Lexis diagrams can for example be analyzed by counting markings, such as dots (30) or lines, across a section of the diagram (32), to identify patterns.’
On line 215-219: ‘The process owners analyzed data as they saw fit in order to find relevant patterns. This included eye-balling the graph for overall trends, counting dots and lines, or analyzing sections of the diagram. It was clarified to process owners that the patterns may indicate trends but that this does not imply statistical significance.’

No comparison is made to the statistical survival curve in this updated draft.

Line 257 - "no clear trend seen when all data included" - I'm confused as to whether this means Figure 3b is wrong or just showing something different? Surely if you look at the same data in different ways and get different answers that is problematic? Again do you mean just by eyeballing the data? or counting the dots?

Author response: Figure 3b is deleted.

Line 266 - what is meant by the "number of relapses is steady over the years" - how do you know that? Are you counting dots in segments? Then you need to make this clear methodologically?

Author response: The part about relapses is taken away, since Figure 3b was deleted. The methods used in data interpretation has been clarified generally on line 125-127: ‘Lexis diagrams can for example be analyzed by counting markings, such as dots (30) or lines, across a section of the diagram (32), to identify patterns. ‘ and on line 215-217: ‘The process owners analyzed data as they saw fit in order to find relevant patterns. This included eye-balling the graph for overall trends, counting dots and lines, or analyzing sections of the diagram.’

It is also clarified in relation to the figures. For example, regarding Figure 3 (line 241-244): ‘From eye-balling the data, there seem not to be any clear trends regarding survival. However, one important and positive conclusion is that this cohort has no treatment-complication-related deaths (which would have been represented by light green dots).’ See also my answer to your next comment below.

Line 270 - how many green dots would signify a problem? Seems a vague statement and is not clear how this would link to action.

Author response: The revised text is (line 277-285): ‘For example, counting light green dots revealed that in the cohort diagnosed during the first four years, 2008–2011, there were five deaths due to treatment, but only one in the cohort diagnosed in 2012 and onwards, covering five years. As noted by a process owner, increase in green dots in Figure 4 during real-time follow-up would represent a timely alarm that there is a problem with the care process. Generally, Lexis diagrams do not rely on any specific rules or thresholds for when to act; rather, it is driven by domain expertise. As treatment-related death should preferably never happen, a single green dot could be enough to trigger further investigation and action.’
Line 325 - I think the point about the two methods being complementary should be made much earlier - i.e. in the introduction when explaining the different methods

Author response: The point about methods being complementary is now made already on line 108: ‘Lexis diagrams are one such as possible method, used to identify joint effects of age, period, and cohorts (24, 25), and serve as a possible complement to subsequent statistical survival analysis (26).’

Line 342 - how was the process owners perception confirmed?

Author response: This sentence is removed, based on a comment from reviewer 2.

Line 350 - 357 - is this a suggestion for future work? Should this belong in the discussion?

Author response: This part has been deleted since the paper was considered (too) long (comment 3 from reviewer 3) and this part is not central for the paper.

Line 392 - how do you know may deaths are due to leiomyoscaroma? Dot counting?

Author response: Yes, the text now explains (line 400-401): ‘Different-colored lines in Figure 6 show the distribution of different sarcoma types, where the lines or dots representing deaths per sarcoma type were counted.’

Discussion

I'm still not clear on how the Lexis diagram aids prediction - could some clear examples be provided of the role it can play here? How could the prediction be tested?

Author response: Thank you for pointing that out! I agree that prediction may be difficult to achieve with Lexis diagram, the focus has been changed to support understanding through feedback by following changes over time.

Line 436 - I am not sure it is within the scope of this paper to make recommendations for how data coding issues should be resolved - this has not been the focus of the paper so this section should be only focus on known limitations and suggestions for future work (not unrelated recommendations).

Author response: This part has been deleted.
The discussion would benefit from a comparison with other QI data methods - especially given the role of control charts is raised (and dismissed) in the introduction it would be helpful to make clear the benefits/limitations of each approach here.

Author response: The three methods control chart, Kaplan-Meier survival curves and Lexis diagrams are now compared in Table 1.

I also wonder if most of the visualisation interpretations you discuss could not just be found with an excel spread sheet and an interested clinician looking at the data and trends? I assume a value of these charts is that not many clinicians will sit and analyse an excel spreadsheet - and so here you are presenting something that is visually intriguing that allows new ideas and interpretations to come to mind that can then be further explored (by counting the dots?? or more advanced methods?). It would be helpful for this to be clearer.

Author response: I have raised this issue in the discussion (line 422-424): ‘Data visualization seemed to be an important driver, as some analysis, such as deaths due to sarcomas, could have been analyzed in tabular form also, but data visualization was still preferred by most process owners, reflecting the power of visualization (49).’

Niels Keiding, Prof. (Reviewer 2): This manuscript describes the use of Lexis diagrams to illustrate medical follow-up studies. The author makes several useful points although reference to previous analyses of one of the data examples could be more precise. The Lexis diagram material is in some sense a direct follow-up of the golden age of the Lexis diagram in Germany 1870-80, see e.g. Keiding (2011) and the pretty coloured Lexis diagrams of Perozzo (1880).

My comments are intended to help make the message become more precise, more clearly explained and better accepted.

Author response: Thank you very much for constructive criticism, pointing out key improvement possibilities of the paper! I highly appreciate the time and effort you have put into the review.

1. The exploratory Lexis diagrams and the confirmatory statistical analyses and associated graphs.

For much of the paper a conflict is postulated between exploratory Lexis diagrams (the good boys) and the confirmatory statistical analyses and associated graphs (the bad or at least dull boys). This distinction is not conducive to what the main message should be: that both are necessary but they fulfill separate purposes. Please modify in this direction.

Author response: The introduction is heavily revised and the comparison between the methods have been reduced in favor of highlighting their different purposes and complementary use.
For example, on line 54-62: ‘As process variation is constantly affected by a wide range of factors, both within and outside of the practitioners’ control (3), feedback gained by following data over time is needed to enable an understanding of the process (4), preferably in real time (5). Data visualization enables analysis and sensemaking by domain experts based on their knowledge and experience (6, 7). They identify and observe datapoints, discover patterns, and compare the results to their prior understanding, drawing inferences and building hypotheses (8). This may answer questions such as “Are we on track?” “Is something negative happening that we need to address?” or even “Is the process improving?” Such exploration may later be followed by confirmatory statistical analysis (9).’

Later, on line 68-73: ‘However, one area in which improvements could be made in the visual representation of data for QI purposes is survival data analysis. Such data are often analyzed through survival curves and used for evaluating improvement efforts (see e.g. 14), but survival curves have another purpose; they contribute to “global knowledge” useful in (e.g.) medical research such as clinical trials of new treatments, or, as in Dahm-Kährler et al. (15), showing that centralization of surgery improves survival on a population level.’

And on line 78-82: ‘For further information about Kaplan–Meier curves, see Jager et al. (16). Its purpose is thus not to support local process understanding and action for QI and due to the need for large sample sizes and aggregation of data, other methods are needed to ensure timeliness and gaining feedback by following data over time, contributing to local improvements.’

2. The overoptimistic management jargon

I was put a little off by the description of the collaboration with medical researchers underlying the story in the manuscript. To me figures such as Fig. 2 are unhelpful and irritating. The collaboration with many meetings between statistician and subject matter researchers and gradual development and understanding of the meaning of the tools is hopefully nothing unique in the author's experience. For me and my group this is how we work all the time - to write in such detail about the number of meetings etc. indirectly raises a suspicion that this was a rare experience in the author's world. And there are too many quotes from these Swedish doctors - for a scientific journal it is not so important what they say but what the author with his/her competence ends up recommending.

Author response: I see your point, thank you! This text is included as part of presenting the qualitative aspects of the study (see e.g. Flick, U. (2014). An introduction to qualitative research. Thousand Oaks, CA: Sage.). Similar text has also been used in this journal before (see e.g. https://doi.org/10.1186/1472-6947-14-60). I have however shortened the text to address this comment.

3. The layout of the Lexis diagrams, the terrible legends

The Lexis diagrams are too small, very difficult to read, this being a problem so serious that I may have missed important parts of the message. I am certain that our big idol Edward Tufte
would disapprove. If the journal requests so small and unreadable graphs, you will have to consider finding a different journal. And I am totally sure that Tufte would never accept writing in big black letters 'red dots' instead of showing a dot in red colour. All symbols in the legend have to be shown as they look on the graph, never black letters with verbal descriptions!!

Author response: Thank you for pointing this out. I have updated the diagram legends and improved the visualizations through increasing the resolution, which also enables increasing readability by zooming the visualization on the computer. As I am aware that some figures may be difficult to interpret, I have added a note in the figure text for figure 4, encouraging readers to use the computer for better display.

It is clear that this ambition of showing all individual lines can only work for rather small data sets, which should be part of the Discussion, if possible with suggestions as to what could be done with larger (more realistic) data sets.

Author response: This issue is now raised in the discussion: (line 437-439) ‘Basic Lexis diagrams are mainly useful for small data sets (30), and large data sets should preferably be analyzed using more advanced Lexis diagrams (26, 34).’

4. Ovarian cancer

I looked up the paper by Damm-Kähler et al. (2016) and they are comparing the cohorts 2008-2010 with 2011-2013 using relative mortality concepts that readers of the present manuscript get no information about. End of follow-up was for both cohorts end of 2015 and median follow-up was 59 months and 41 months, respectively (see top of their p. 213). It is therefore incorrect to claim, as the present author does on p. 10, that 'these patients are followed for only three years'. Maybe the present author did not understand what Damm-Kähler et al. did? Since patients who died after three years but still within the original follow-up are classified as survivors in the Lexis diagram shown in Fig.3b, this is not a fair comparison to the plot from Damm-Kähler et al. in Fig. 3a. It would have been more pedagogic to display the relevant Lexis diagram for the complete follow-up underlying the analyses in the paper by Damm-Kähler et al. As the later Fig. 4, this would have a triangular shape, as these very often have, see Axelsson et al. (2019a,b) for examples.

Author response: Thank you for this clarification! The comparison between the survival curves and Lexis diagram has been deleted, along with figure 3b.

Fig. 3

a. What do the red dots at the zero line mean? On p. 11 line 8 the author writes

Each dot in the Lexis diagram within a time interval…..represents the number of deaths
I wonder what the author means? Does one dot represent 8 deaths, another dot 25 deaths, or do all dots represent 1 death?? In any case it is so difficult to see how many dots there are that some clarification is necessary.

Author response: This is a writing error, one dot represents one death. The text has been updated (line 239-240): ‘Each dot represents one death, and the different colors of death events represent causes of death, as presented in the legend.’

b. I am not sure the downward lines are such a good idea, and one wonders what happens to patients who are known to die but more than three years after diagnosis. As far as I can see they have just disappeared. No downward line since dead at data extraction, no upward line since not dead at 3 years after diagnosis.

Author response: The figure with downwards lines is deleted, partly based on your comment nr 4.

c. In the description of Fig. 3 on page 10 we learn that 'as patients decease, their lifelines move from going downwards to upwards graph' - but in which sophisticated dynamical space does this take place? I am afraid you are losing your target group here.

Author response: The figure is deleted.

Fig. 4

Again: instead of writing 'Blue' in black letters, show us a blue line in blue, and so on.

Author response: The legends have been changed accordingly.

In the text on p.11, line247-248, it says

'Instead of restricting analysis to 3-year survival as survival curves do'  
But somebody will have to tell the survival curves to cover whatever time range desired. So the 3 years are of course a choice of the analyst, as are the choices in the Lexis diagram. More importantly, this passage sets up a competition between survival analysis and Lexis diagrams, which, as I explained above, is totally misplaced: these two tools supplement each other, should not be seen as competitors.

Author response: My mistake. The figure is deleted and although some comparison between the methods are made by the process owners, I have tried to emphasize that there is no competition between the methods, rather differences in purposes.
Cohorts and calendar time: please be more careful when talking about years, e.g. 2012-14: do you mean events in those calendar years or for the cohorts diagnosed in those years. Do not forget that the original purpose (and still the most important purpose) of Lexis diagrams is to keep track of the joint effects of age, period and cohort. An important example is p. 12, lines 266 ff.:

Although the number of relapses is steady over the years, we see fewer deaths in the first year, especially from early 2014 where the meaning (hopefully) is the cohorts, not calendar year - but will the uninitiated reader catch this?

Author response: The difference between time and cohorts has been clarified, for example in lines 277-280: ‘For example, counting light green dots revealed that in the cohort diagnosed during the first four years, 2008–2011, there were five deaths due to treatment, but only one in the cohort diagnosed in 2012 and onwards, covering five years.’

Cervical cancer

The initial statement

In survival curves, calendar time is the only time axis available for visual presentation

Is not true for experienced survival analysts - although it is true that the Lexis diagram is a useful additional tool.

Author response: The text is revised to (line 346-347): ‘One strength of Lexis diagrams is to present data along several time axes. In this example, age is used on an additional time axis to diagnosis date time axis.’ This also reduces the comparison/competition between methods.

At the bottom of page 15 we learn that 'mainly older patients died'. Not too surprising, even if one is not a Swedish M.D. The case-by-case remarks in that section run the danger of being misunderstood as hard evidence by uninitiated readers. Please make sure that serious statistical analysis confirms the intuitive impression.

Author response: The text is revised to avoid misunderstanding (line 358-363): ‘Contradicting the process owner’s perceived clinical experience, it appeared that age seemed to have had little to do with the deceased’s length of survival in this dataset; indeed, two of three patients who were 90 or older survived for several years and one of them died tumor free from another cause. As with any trend identified, however, such relations need to be tested using proper statistical principles (46) before sound conclusions can be drawn.’

References


Author response: This reference has been cited (nr 26).


Author response: This reference has been cited (nr 25).


Ralph Brinks, Ph.D. (Reviewer 3): Summary: The paper suggests the Lexis diagram as a tool for data visualization in chronic conditions. The suggestion is illustrated by data about cancer in Sweden. The topic is important and the paper contributes to the discussion. However, in some parts it should be written more concise and clearer.

Author response: Thank you for helpful comments and the time you have spent on reviewing the paper!

Major points:

1. Background: The first paragraph after the epigraph has many breaks in contents. It jumps around quality, data, statistical significance, prediction, system behavior and graphical displays. All of these are important points, partly controversially discussed (see the recent debate about the p-value). However, I would recommend to write a few more sentences to bring these aspects together. In its current form, the reader will hardly be able to follow what is exactly meant.

Author response: Much of the background is revised to address this comment as well as several comments from Reviewer 1 and 2. The revised text in the first paragraph is (line 50-65): ‘The aim of quality improvement (QI) is “to make changes that will lead to better patient outcomes (health), better systems performance (care) and better professional development (learning)” (1, p. 2). Improvement efforts should be guided by data, which can enable practitioners to understand how their local processes vary and take relevant action on that basis (2, 3). As process variation
is constantly affected by a wide range of factors, both within and outside of the practitioners’ control (3), feedback gained by following data over time is needed to enable an understanding of the process (4), preferably in real time (5). Data visualization enables analysis and sensemaking by domain experts based on their knowledge and experience (6, 7). They identify and observe datapoints, discover patterns, and compare the results to their prior understanding, drawing inferences and building hypotheses (8). This may answer questions such as “Are we on track?” “Is something negative happening that we need to address?” “or even “Is the process improving?” Such exploration may later be followed by confirmatory statistical analysis (9). Data visualization also has the purpose of communicating findings (10), and has been used as a basis for discussion between stakeholders for QI (11). Performance-related feedback has also been shown to be important for motivating staff, regardless of whether it indicates a need for improvement or not (12).

I kindly refer to the submitted updated draft for reading the rest of the background.

2. For some chronic conditions, more than two time scales are important. For instance, Bendix Carstensen emphasis that, apart from age and period, duration of diabetes plays an important role for mortality. In this case, the idea of the Lexis diagram can be extended to a three dimensional visualization (see DOI 10.1371/journal.pone.0106043). Then, a simple planar representation as in Fig 1, 3b, 4-6 of the manuscript is not possible anymore. It remains open if a planar projection is capable to represent duration. A similar problem occurs in remission and relapse of cancer. I ask myself if in a graph like Fig 4 it is possible to assess if time to remission becomes shorter and/or time to relapse becomes longer. Due to medical progress (better treatment, better care), we would expect secular trends there. These aspects as well as dependency on the duration of the chronic condition should be discussed critically with regard to the visualization by planar Lexis diagrams.

Author response: Thank you for this input! The discussion now includes this as a major drawback of basic Lexis diagrams:461-465) “One potential drawback with the basic Lexis diagram is the possibility that intermediate events, such as relapse, may affect how survival or the duration of the disease is dependent on time (53). In such a case, an additional time axis in the Lexis diagram could be used, rendering a 3D visualization (54).”, where reference nr 54 is your suggested paper with DOI 10.1371/journal.pone.0106043.

Later, this reference is also included in suggestion for future research on interactive Lexis diagrams, line 478-482): ‘ …future research may strive to develop interactive Lexis diagram platforms, enabling attribute data to be chosen depending on need and helping viewers understand the relatedness of clinically relevant attribute datapoints (23). These platforms could be extended to include 3D visualization as well, to address secular time trends (54). ‘

Minor points:
1. It appears unusual that the Background section of a scientific article starts with an epigraph. In my opinion, this is more appropriate in book chapters. Thus, I recommend to remove the quotation.

Author response: The quotation is moved to a later part of the background (line 108-110).

2. The numbers of ordinate axis ("y-axis") in Fig 1 and Fig 3b should be rotated by 90 degree clockwise for better readability. In the used statistical software R, the command is "las = 1".

Author response: Thank you for your kind help! This has been done.

3. The manuscript is quite long. I think parts of it (e.g. the quotations of the process owners) can be put as supplementary information into the web appendix.

Author response: I agree that the manuscript is quite long and I have removed some quotations, as well as some other parts of the original draft. I am however reluctant to put the quotes as supplementary information as they represent the main results of the paper and clarify the discussion and conclusion. They are also in line with Reviewer 1, being interested in the practical use of data (and the need to appropriately visualize complex information) to drive action. The quotes explain the process owners’ potential practical use of the data/Lexis diagrams and actions they may take based on the insights they get.