

## PEER REVIEW HISTORY

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### ARTICLE DETAILS

<b>TITLE (PROVISIONAL)</b>	Funnel plots and choropleth maps in cancer risk communication: a comparison of tools for disseminating population-based incidence data to stakeholders.
<b>AUTHORS</b>	MAZZUCCO, WALTER; Cusimano, Rosanna; Zarcone, Maurizio; Mazzola, Sergio; Vitale, Francesco

### VERSION 1 - REVIEW

<b>REVIEWER</b>	Bin Zhang Cincinnati Children's Hospital Medical Center USA
<b>REVIEW RETURNED</b>	05-May-2016

<b>GENERAL COMMENTS</b>	The paper was well written. But the results need more interpretation.
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<b>REVIEWER</b>	Bradley Manktelow University of Leicester United Kingdom
<b>REVIEW RETURNED</b>	23-May-2016

<b>GENERAL COMMENTS</b>	<p>The authors propose the use of Funnel Plots in order to display epidemiologic cancer data, using information from the Palermo Province Cancer Registry as an example.</p> <p>The authors state that the aim of this paper "... is to propose the use of funnel plot ..." However, in my opinion more detail and discussion is required in order for readers to make an informed choice of methodology. In addition, the example chosen seems to present specific challenges in that there is one area which includes a large proportion of the whole population included in the paper.</p> <p>Major comments</p> <p>1) As the authors have pointed out, the proposed use of funnel plots to display such data is not new and the first papers proposing their use for observational data are now over 10 years old. Although I am not aware of the previous use of funnel plots with data from population based cancer registries, it would be helpful if the authors could more clearly state what is novel about this application and how it differs from previous applications.</p> <p>2) The authors do not discuss any of the potential disadvantages with the use of funnel plot. For example, the issue of the value of the type I error not matching the nominal error rate (e.g. 99.8%) especially when the benchmark is obtained from the data – as it is in</p>
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	<p>the example shown. A more complete discussion of the advantages and disadvantages of funnel plots should be included since the authors are recommending their use and readers should be able to make an informed choice.</p> <p>3) The authors state that "...understanding of the confidence interval is poor..." I think that the authors should offer a description and discussion of the funnel plot control limits (confidence intervals? prediction intervals?) to explain what they are and their characteristics. It is not clear to me how someone whose understanding of confidence intervals is poor would have a better understanding of the correct interpretation of funnel plots. Without more detailed explanation there is a danger of the methodology being a 'black box' with users having insufficient knowledge to interpret them correctly.</p> <p>4) In the example shown, the potential outlying area (Palermo City) seems very different from the other areas in terms of population size. One potential problem with this is that this area has a major impact on the estimation of the value of the line of no difference. One effect of this is that the other areas almost all fall below the line of no effect. This raises the question of whether the underlying hypothesis underlying the funnel plot is plausible in this case. This hypothesis is that the underlying mortality within each area is identical to that of the whole cohort. The authors should consider and discuss the assumptions underlying the use of funnel plots, with particular consideration of their example where one area has such a large proportion of the population.</p> <p>Minor comments</p> <p>5) The authors state that extra information can be obtained by email. Could this be made available as an appendix? This information would be helpful to the readers as details are lacking in the paper.</p> <p>6) There are some minor issues with the use of English which need correcting.</p>
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<b>REVIEWER</b>	Ariana Znaor International Agency for Research on Cancer, France
<b>REVIEW RETURNED</b>	18-Jun-2016

<b>GENERAL COMMENTS</b>	<p>Exploring ways to more effectively communicate epidemiological data to stakeholders is of great importance, and funnel plots have already been successfully used to communicate data from other areas of cancer surveillance. However, in addition to several comments listed below, I would suggest to postpone publication until the results have been validated in the field as planned, which would make the article more informative.</p> <p>General: The article would benefit of careful editing, some sentences remain unclear and several references are used incorrectly (eg. ref no 8 on cancer trends in NW Iran is used to document global population aging).</p> <p>Background: End of 1st paragraph: The statement on public health need for more</p>
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	<p>information on cancer causes might mislead the reader to think that PBCR data presented by funnel plots would provide those.  2nd paragraph: To describe the role of PBCRs international references should be used rather than regional reports (refs 13,14). Moreover, the statement that PBCRs continuously record and monitor cancer incidence, mortality, survival and prevalence is inaccurate. Many PBCRs worldwide do not have access to mortality data, whereas many others use them as a data source, but are not in charge of recording and monitoring cancer mortality.</p> <p>Methods:  The indirect standardization method is not described in enough detail. Reference population is not specified (presumably the total population of PP, but this should be listed and also added to the table). Even though the article focuses on data presentation rather than results themselves, it would be worthwhile to present the results separately for men and for women, as geographical patterns might substantially differ by sex.</p>
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### VERSION 1 – AUTHOR RESPONSE

Point-by-point reply to Reviewer: 1

Reviewer Name Bin Zhang Institution and Country Cincinnati Children's Hospital Medical Center USA

COMMENT: - The paper was well written. But the results need more interpretation.

REPLY: Authors thanks the reviewer for the comments. The discussion has been implemented with further interpretations, also according to additional results and supplementary material provided in the manuscript.

Point-by-point reply to Reviewer: 2

Reviewer Name Bradley Manktelow Institution and Country University of Leicester United Kingdom

COMMENT: The authors propose the use of Funnel Plots in order to display epidemiologic cancer data, using information from the Palermo Province Cancer Registry as an example. The authors state that the aim of this paper "... is to propose the use of funnel plot ...". However, in my opinion more detail and discussion is required in order for readers to make an informed choice of methodology. In addition, the example chosen seems to present specific challenges in that there is one area which includes a large proportion of the whole population included in the paper.

REPLY: Authors thanks the reviewer for the useful comments. The discussion has been further implemented with more details and considerations according to additional results and to supplementary material provided. We believe that the revisions will help the readers to make a more informed choice of the proposed methodology.

With regard to the example chosen, as remarked by the reviewer, it is true that the presence of "one area which includes a large proportion of the whole population included in the paper" could affect the results, but this effect is related to the reference population used in the calculation of SIRs and not to FP methodology itself. Moreover, we believe that use of FP, which allows to display the position of a single municipality in relation to the entire province, is much more informative about the meaning of the punctual estimates (SIRs) and their CIs rather than the same numerical values reported in a table. This is also true independently from the administrative characteristics of the study population, even for the one considered in the proposed example, documenting a large proportion of the entire population in a specific area. It has also to be considered that the SIRs' values have been standardised having the EU population as external reference.

We have further argued in reply to COMMENT 4.

## Major comments

COMMENT 1: As the authors have pointed out, the proposed use of funnel plots to display such data is not new and the first papers proposing their use for observational data are now over 10 years old. Although I am not aware of the previous use of funnel plots with data from population based cancer registries, it would be helpful if the authors could more clearly state what is novel about this application and how it differs from previous applications.

REPLY 1: What was explored for the first time in our paper is the use of funnel plot to represent a measure of incidence (standardized incidence ratio), whereas the previous applications of FP by population-based cancer registry almost referred to survival and to mortality measures.

- Consequently, we have modified the Discussion text as follows: (Page 15, line 9): "FP are commonly used in process control and, in particular, in the health care field to compare institutional performance data; however, this format is used for survival and standardized mortality ratio in public health surveillance."

- We have also modified the Background section as follows: (Page 8, line 34) "The aim of this paper is to propose the use of funnel plots (FPs) for reporting local cancer incidence data, as an alternative to the more common visual formats employed by the PPCR to address local public health authorities and communities, in order to facilitate the dissemination and interpretation of measures of cancer statistics at the municipal level."

- Lastly, we have updated the Conclusion section as follows: (Page 17, line 52) "According to the proposed comparison between the two explored methodological approaches, we concluded that FP should be considered as an alternative to the current and commonly used graphical and visual formats (CMs, tables, GAM maps) to effectively communicate cancer registry statistics, particularly incidence rate, to communities and local authorities, visually conveying an efficient and simple to interpret cancer epidemiological data."

COMMENT 2: The authors do not discuss any of the potential disadvantages with the use of funnel plot. A more complete discussion of the advantages and disadvantages of funnel plots should be included since the authors are recommending their use and readers should be able to make an informed choice.

REPLY 2: - We have implemented the Discussion section as follows, according to what is presented in Table 2: (Page 17, line 9): "In particular, with regard to the strengths of the proposed visual format, FP shows the scope of the phenomenon under investigation and the precision and significance of estimates simultaneously, by simply positioning the indicator of interest in one of the three cancer attention areas; on the contrary, the more commonly used CMs monodimensionally represent the parameters of interest by using a different color gradation based on the frequency distribution of the values. The highlighted difference could be considered the main reason for making FP more comprehensive to stakeholders than CM. However, the weaknesses of FP also need to be taken into account. FP cannot be considered the ideal visual format to highlight the geographical position of the indicator of interest (SIR) and, consequently, to define any spatial cluster. Lastly, both FP and CM had the ability to identify potential hot spots, even though for CM, it is necessary to further validate the hot spot by using suitable statistical tests (for example the GAM approach). All of the previous considerations have led us to believe that FP could be preferable to CM both in terms of validity and in terms of interpretability."

- We have also modified the Results section as follows: (Page 13, line 37) "Table 2 summarizes a comparison of the weaknesses and strengths, as per the available literature, between the different visual formats explored within the context of disseminating epidemiological data to stakeholders."

COMMENT 2B: For example, the issue of the value of the type I error not matching the nominal error rate (e.g. 99.8%) especially when the benchmark is obtained from the data – as it is in the example shown.

REPLY 2B: Concerning the proposed example, the SIRs have been calculated over a period of 9 years, involving a large number of observed cases and of person-years in the calculation of the

expected cases. Therefore, we were able to calculate the confidence intervals of each SIR by using the normal approximation instead of using the Poisson distribution. Also, the use of the normal distribution make us possible to calculate the exactly type I error. Moreover, to avoid some values to erroneously fall in an “out of control” area, the overdispersion has been taken into account. Lastly, the confidence levels (5% and 0,2%) used to develop our model are the same applied to FP’s standard methodology [Spiegerhalter et al.].

COMMENT 3: The authors state that “...understanding of the confidence interval is poor...” I think that the authors should offer a description and discussion of the funnel plot control limits (confidence intervals? prediction intervals?) to explain what they are and their characteristics. It is not clear to me how someone whose understanding of confidence intervals is poor would have a better understanding of the correct interpretation of funnel plots. Without more detailed explanation there is a danger of the methodology being a ‘black box’ with users having insufficient knowledge to interpret them correctly.

REPLY 3: We agree with the reviewer. That is why we believe that it is easier for a reader “not familiar with statistics and epidemiology” to get the message coming from a point (SIR of the municipality of residence) situated within the different attention areas of the graphic, defined by different colors, than the numerical representation of CIs.

- Therefore, we have modified Figure 1, by splitting it in two different graphs: (Page 28) “Figure 1. a) Funnel plot of the SIRs in the 82 Palermo Province municipalities (Study period 2003-2011); b) Cancer attention areas: “under-control” area (in green), “warning” area (in yellow) and “alert” area (in red).”

- We have also implemented:

-) the Methods section with the following sentence: (Page 9, Line 51) “The two sets of control limit lines define three different areas within the graph (Figure 1b): the “under-control” area (in green), the “warning” area (in yellow) and the “alert” area (in red).”

-) the Results section as follows: (Page 12, line 10) “Figure 1a represents the FP of 82 municipality-specific SIRs, corrected for overdispersion ( $\phi = 13.46$ ) and adjusted using the multiplicative approach. All of the SIRs lay within the control limits, except for the Palermo city one (SIR= 1.12), which resulted above the upper control limit line (UCL) of 99.8%. Figure 1b identifies the three different cancer risk areas within the graph.”

-) the Discussions section as follows: (Page 15, Line 47) “Therefore, in order to facilitate comprehension of the epidemiological message, we have chosen the FP as a visual display method to allow the reader to identify the SIR for each municipality within the plot, and the different attention level areas (represented by different colors) under which each location falls (Figure 1b).”

COMMENT 4: In the example shown, the potential outlying area (Palermo City) seems very different from the other areas in terms of population size. One potential problem with this is that this area has a major impact on the estimation of the value of the line of no difference. One effect of this is that the other areas almost all fall below the line of no effect. This raises the question of whether the underlying hypothesis underlying the funnel plot is plausible in this case. This hypothesis is that the underlying mortality within each area is identical to that of the whole cohort. The authors should consider and discuss the assumptions underlying the use of funnel plots, with particular consideration of their example where one area has such a large proportion of the population.

REPLY 4: As reminded above, the purpose of this article is to facilitate the comprehension of the readers on the epidemiological data (cancer burden) coming from population-based cancer registries. The FP graphical representation, differently from the commonly used visual formats, allows the reader to simply display at the same time the position of his municipality with respect to the entire reference population and to three potential different areas (under-control, warning and alert) representing the different levels of attention.

Getting to the proposed example, it is true that the presence of an area with a very large population (Palermo City) implies an overestimation of the expected cases. However, we have further performed

the analysis excluding Palermo city and we have observed that the message deriving from the “visual” representation didn’t change.

Consequently, we have implemented the Discussion section as follows: (Page 16, line 25) “Within the context of the chosen sample population and data, it has to be considered the presence of a single area containing a large proportion of the entire study population must be highlighted. This obviously influences each SIR value, but its potential effects are related to the study population used in the calculation of SIRs, and do not influence the FP methodology itself. Moreover, the graphic FP representation, differently from the more commonly used visual formats, allows the reader to observe, simultaneously, the situation of the municipality of interest in relation to the entire study population and to three specific areas (under control, warning and alert) representing the different attention levels. Moreover, it should also be kept in mind that the SIR values have been standardised using the EU population as external reference, allowing adjustment for age. Lastly, the presence of a single area with a substantial population (Palermo city) implies an overestimation of expected cases, but the epidemiological message did not change even after the exclusion of the Palermo city area from the analysis (data not shown).”

#### Minor comments

COMMENT 5: The authors state that extra information can be obtained by email. Could this be made available as an appendix? This information would be helpful to the readers as details are lacking in the paper.

REPLY 5: We have attached a supplementary file as suggested by the reviewer.

- We have cited it in the Methods section (page 10, line 21): “Moreover, Z-score and the winsorization method (by testing for different levels of Z-score quantiles) were applied for the direct selection of extreme values. Furthermore, to define the level of winsorization, an R-script routine was developed to set a cut-off for the quantile between the acceptance and rejection of the overdispersion test (Supplementary material).”

- We have also modified the Data sharing statement as follows: (Page 19) “Supplementary data (results of over-dispersion tests, R-script to detect the greatest cut-off for the winsorization procedure) have been provided as a supplementary file. Other statistical results are available by emailing [walter.mazzucco@unipa.it](mailto:walter.mazzucco@unipa.it).”

COMMENT 6: There are some minor issues with the use of English which need correcting.

REPLY 6: The manuscript has been reviewed and corrected by a native English speaker with extensive scientific editorial experience to ensure a high level of spelling, grammar and punctuation. (See the acknowledgement section).

Point-by-point reply to Reviewer: 3

Reviewer Name Ariana Znaor Institution and Country International Agency for Research on Cancer, France

COMMENT: - Exploring ways to more effectively communicate epidemiological data to stakeholders is of great importance, and funnel plots have already been successfully used to communicate data from other areas of cancer surveillance. However, in addition to several comments listed below, I would suggest to postpone publication until the results have been validated in the field as planned, which would make the article more informative.

REPLY: We thank the reviewer for the very useful comments.

- We have recalled along the Discussion section all of the FP documented applications, including the one for cancer surveillance: (Page 15, line 9): “FP are commonly used in process control and, in particular, in the health care field to compare institutional performance data; however, this format is used for survival and standardized mortality ratio in public health surveillance.”

- Concerning the reviewer suggestion to postpone the manuscript publication until the results have been validated in the field, authors are still confident that the explored results support their

hypotheses.

By the way, in order to perform the validation in the field of the proposed methodological approach, the Palermo Province Cancer Registry team has developed a specific tool, the “municipal cancer report”, and it’ll take us not less than one year to administer it to the 82 PP local communities (citizens, local authorities, general practitioners, specialized physicians, pharmacists, etc.). Therefore, we believe that the further evidences representing the experience “in the field” could be of interest for a future manuscript.

- Anyway, to better disclose the next steps, we have modified the Discussion section as follows: (Page 17, line 36) “However, the proposed alternative dissemination approach needs to be further validated in the field both by involving local communities and by administering the two different visual formats to a sample of stakeholders according to the Delphi consensus process. ”

GENERAL COMMENT: The article would benefit of careful editing, some sentences remain unclear and several references are used incorrectly (eg. ref no 8 on cancer trends in NW Iran is used to document global population aging).

REPLY TO GENERAL COMMENT: Authors thank again the reviewer for the suggestions.

- An editing of the text has been provided. The manuscript has also been reviewed and corrected by a native English speaker with extensive scientific editorial experience to ensure a high level of spelling, grammar and punctuation. (See the acknowledgement section).

- Further, references have been reviewed and reference no.8 has been changed with the following one: WHO Global Status Report on Non-Communicable Diseases 2010. Geneva 2011.

Background:

COMMENT 1: - End of 1st paragraph: The statement on public health need for more information on cancer causes might mislead the reader to think that PBCR data presented by funnel plots would provide those.

REPLY 1: We have modified the Background section as follows: (Page 7, Line 24): “Local public health and political authorities regularly engage in finding better ways to satisfy the growing demand for information on the impact of cancer by the general public. In particular, citizens often question if they live in an area at high risk for environmental exposure.”

COMMENT 2: - 2nd paragraph: To describe the role of PBCRs international references should be used rather than regional reports (refs 13,14). Moreover, the statement that PBCRs continuously record and monitor cancer incidence, mortality, survival and prevalence is inaccurate. Many PBCRs worldwide do not have access to mortality data, whereas many others use them as a data source, but are not in charge of recording and monitoring cancer mortality.

REPLY 2: - We have modified the Background section [and the corresponding references] as follows: (Page 7, line 39) “Population-based cancer registries (PBCRs) carry out cancer surveillance by continuously collecting and classifying information on all new cancer cases within a defined population, and providing statistics on its occurrence for the purpose of assessing and controlling the impact of this disease on the community.”

- Again, we have modified the Background section [and the corresponding references] as follows: (Page 7, line 54) “PBCR publications use validated and internationally shared measurements systems and employ terminology and visual formats that are easily understood by the scientific community, but often difficult to interpret for other stakeholders, particularly at the local level.”

Methods:

COMMENT 3: - The indirect standardization method is not described in enough detail. Reference population is not specified (presumably the total population of PP, but this should be listed and also added to the table).

REPLY 3: - We have modified the Methods section as follows: (Page 9, line 10) “The study population

consists of the 51,951 new cancer cases, excluding non-melanoma skin cancers, registered between 2003 and 2011 by the PPCR among the 1.244.239 residents of the 82 municipalities of the Palermo Province (PP) (679.850 inhabitants within the Palermo metropolitan area only). Cancer incidence in the PP municipalities was measured by using Standardized Incidence Ratio (SIR), defined as the ratio between observed cases ( $O_i$ ) and expected cases ( $E_i$ ). The  $O_i$  were assumed to follow a homogeneous Poisson distribution with parameter  $\lambda = \theta \cdot E_i$ . The  $E_i$  were estimated by indirect method, considering the entire population-time under study (the PP) as the reference population, with  $\sum O_i = \sum E_i$ . The resident population was reported using the inter-census estimates, provided by the Italian National Statistical Institute (ISTAT), also considering the annual municipal data on migration.”

- We have also modified the Table 1 title as follows: (Page 12, line 37) “Table 1. Expected cases and SIRs (shown in a descending order) with 95% CIs in the 82 Palermo Province municipalities (study period: 2003-2011, reference: the entire PP population).”

COMMENT 4: - Even though the article focuses on data presentation rather than results themselves, it would be worthwhile to present the results separately for men and for women, as geographical patterns might substantially differ by sex.

REPLY 4: As the aim of the study is to facilitate the comprehension of the reader on the epidemiological message of cancer incidence by exploring the use of alternative graphical representation – and since the primary interest coming from citizens is to ascertain whether the place of residence implies an high risk of cancer occurrence for the local community - we did not present results by gender. Anyway, answering to the reviewer comment, we can report that in all of 82 explored municipalities the resident populations have a stable male/female ratio. Of course, for further studies implementing the proposed graphical visual format to represent the cancer incidence by tumor site, we plan to use FP stratifying the incidence by sex, as gender differences can be of interest according to the different types of cancer.

#### VERSION 2 – REVIEW

<b>REVIEWER</b>	Bin Zhang CCHMC, USA
<b>REVIEW RETURNED</b>	08-Sep-2016

<b>GENERAL COMMENTS</b>	<p>The authors added the limitation and explained the results in more details. However, there are still several major concerns.</p> <ol style="list-style-type: none"> <li>1. The innovation of this application is not big enough.</li> <li>2. Funnel plot may serve as a supplementation of choropleth maps not a substitution since choropleth maps has a big advantage in presenting the spatial locations which is crucial in public health.</li> <li>3. We all know the CI is just statistical concepts, it depends on a lot of things. Sometimes if we just look at the CIs, it will be misleading. For example, in this paper, the highest SIR is from Isnello, the 95% CI is (0.99, 1.45). Although the CI contains 1, it is still marginal significant. It may due to the small population. It may not be appropriate to just say it is under control. People in Isnello may still have a higher risk compared to other cities.</li> </ol> <p>In summary, funnel plot definitely provides information that is not caught by choropleth maps, but the authors should pay much attention to the interpretation.</p>
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## VERSION 2 – AUTHOR RESPONSE

Point-by-point reply to Reviewer: 1

Reviewer Name Bin Zhang Institution and Country Cincinnati Children's Hospital Medical Center USA

COMMENT 1: The innovation of this application is not big enough.

REPLY 1: Although funnel plots (FPs) are an established tool and have been applied to display data on cancer survival [39] and mortality [29], and more broadly in public health surveillance [40], FPs have not been used in the context of cancer risk communication with non-technical stakeholders such as local government authorities and communities. The comparative analysis we present in this paper highlights the utility of FPs, as compared to choropleth maps in terms of interpretability and of validity, to address local public health authorities and communities in order to facilitate the dissemination and interpretation of measures of cancer statistics at the municipal level.

Thus, the innovative aspect of our work is in recognizing that FPs improve cancer risk communication, and provide the audience with a broad framework for evaluating data, improving the interpretability of comparisons and the validity of the conclusions. As the reviewer points out (see below) FPs are complementary to choropleth maps, and can be used effectively to empower local communities and other non-technical stakeholders.

COMMENT 2: Funnel plot may serve as a supplementation of choropleth maps not a substitution since choropleth maps has a big advantage in presenting the spatial locations which is crucial in public health.

REPLY 2: We agree with the reviewer. FP can be considered as an useful and innovative but complementary tool to the common used visual formats, particularly to CM. Table 2 clearly shows how each visual tool addresses complementary issues. We have acknowledged that FPs are a complement, and not an alternative to CMs in the revised paper as follows:

Abstract – Background section: “Aim of this paper is to compare the commonly used visual formats to funnel plots to enable local public health authorities and communities to access valid and understandable cancer incidence data obtained at the municipal level.”

Abstract – Conclusions section: “Funnel plot should be used as a complementary valuable tool to communicate epidemiological data of cancer registries to communities and local authorities, visually conveying an efficient and simple way to interpret cancer incidence data.”

Background section (last sentence): “The aim of this paper is to propose the use of funnel plots (FPs) for reporting local cancer incidence data, as a complement to the more common visual formats employed by the PPCR to address local public health authorities and communities, in order to facilitate the dissemination and interpretation of measures of cancer statistics at the municipal level.”

Discussion section (page 14, second sentence): “We explored the use of FPs as a supplementary tool to local provide authorities and communities with synthetic access to valid and understandable cancer incidence data (SIRs) obtained at the municipal level. ”

Discussion section (page 15, first sentence): “Following the methodological approach proposed, representation of the Palermo Province SIRs through FP seemed to be congruent with CM generated using the same data, with the former resulting more informative dealing with some of the dimensions explored, as shown by the comparisons of the weaknesses and strengths between the two visual formats (Table 2).”

Discussion section (page 16): “All of the previous considerations have led us to believe that FP could be used as a complement to CM, according to its properties, particularly in terms of validity and in terms of interpretability. However, the proposed complementary dissemination approach needs to be further validated in the field both by involving local communities and by administering the two different visual formats to a sample of stakeholders according to the Delphi consensus process.[44]”

Conclusions section (first sentence): “According to the proposed comparison between the two explored methodological approaches, we concluded that FP should be considered as a complement to the current and commonly used graphical and visual formats (CMs, tables, GAM maps) to effectively communicate cancer registry statistics, particularly incidence rate, to communities and local

authorities, visually conveying an efficient and simple to interpret cancer epidemiological data.”

COMMENT 3: We all know the CI is just statistical concepts, it depends on a lot of things. Sometimes if we just look at the CIs, it will be misleading. For example, in this paper, the highest SIR is from Isnello, the 95% CI is (0.99, 1.45). Although the CI contains 1, it is still marginal significant. It may due to the small population. It may not be appropriate to just say it is under control. People in Isnello may still have a higher risk compared to other cities.

REPLY 3: We agree with the reviewer concerning the limits of SIRs calculated on municipalities with a small population. Consequently, according to the reviewer suggestions, we have modified the text as follows:

Results section (Page 11): “Table 1 represents the expected cases (both males and females) and SIRs with 95% CIs in the 82 PP municipalities: most of the SIRs are lower than 1 and only six municipalities present SIRs higher than 1. Among them only Palermo had a statistically significant value higher than 1 (SIR=1,12; 95% CIs= 1.11-1.14) while Isnello, the municipality showing the highest SIR, failed to meet the conventional criteria for statistical significance (SIR=1.22; 95% CIs 0.99-1.45).”

Discussion section (page 14, last sentence, page 15, first sentence): “On the other hand, the conservative choice of reporting only statistically significant increased cancer risks, as shown for the Palermo city hot spot (Figure 3), excludes from the discussion the residents of most municipalities who would certainly be interested in knowing “what is going on in their back yard”. The combination of FP and choropleth map, supported by tabulation of the numeric results, allows to identify locations where cancer incidence may deserves further attention, such as the municipality of Isnello, with a high SIR but a 95% CI including the null value. Clear understanding by the relevant stakeholders and their productive engagement may clarify whether such borderline findings simply reflect inadequate sample size, chance or a departure from the expected incidence that deserves further investigation.

FINAL COMMENT: In summary, funnel plot definitely provides information that is not caught by choropleth maps, but the authors should pay much attention to the interpret

REPLY TO FINAL COMMENT: We thank the reviewer for the very useful comments. According to the comparative analysis, we re-iterate that the FP enhances the reader’s ability to interpret the data and it could be especially helpful for readers who are not trained in statistics, but at the same time are involved in the process because of a perceived risk (citizens) or are engaged in the management of it (local health and political authorities as majors or public health operators or GPs, etc.).

The novelty of this paper lies in showing how FPs can serve as a useful complement to the CM in addressing risk communication with local communities, helping surveillance systems like population-based cancer registries better serve the population and their government authorities.