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Forensic research on sudden death published in Spain☆

Xavier Covas-Cerdà[a], **Ignasi Galtés**[b,c,*]

[a] *Hospital Universitari Parc Taulí de Sabadell, Sabadell, Barcelona, Spain*
[b] *Institut de Medicina Legal i Ciències Forenses de Catalunya (IMLCFC), Barcelona, Spain*
[c] *Unitat de Medicina Legal, Facultat de Medicina, Universitat Autònoma de Barcelona, Barcelona, Spain*

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Abstract
Introduction: One of the main pillars of the work of legal medicine in Spain is research. Taking sudden death (SD), a classical subject in forensic pathology, as the reference this paper aims to examine developments in Spanish forensic research papers published in the two most relevant Spanish legal medicine journals.
Methods: Articles on SD over the last 10 years from the two most relevant Spanish forensic journals were gathered. An international journal with medium-intermedium impact factor (second quartile in 2018) was the comparator. A third open search of PubMed indexed medical journals was conducted to detect and treat potential derivative bias.
Results and discussion: Cardiac SD is the most studied SD at a national level, with high rates of collaboration among centres and specialties. However, this does not result in higher rates of original papers, but in more special articles and case-reports. At an international level, case-reports are the main publication format followed by original articles, mainly focussing on non-cardiac SD.
Conclusions: As a prospect for the future, we suggest strengthening policies to promote research and, above all, to create collaboration networks among specialties towards a multidisciplinary, preventive and clinical approach to SD research.

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* Corresponding author.
E-mail address: ignasigaltes@gmail.com (I. Galtés).

PALABRAS CLAVE
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Investigación forense sobre muerte súbita publicada en España

Resumen
Introducción: Uno de los pilares básicos en la labor de la Medicina Legal y Forense en España es la investigación. Tomando como referencia la muerte súbita, tema clásico de la patología forense, se pretende realizar un estudio del estado de salud de la investigación forense que se publica en las dos revistas españolas más relevantes del ámbito de la medicolegal.
Metodología: Se recogen artículos sobre muerte súbita de los últimos 10 años (2009–2018) de*Revista Española de Medicina Legal* y *Cuadernos de Medicina Forense*. Como comparador, una revista internacional con factor de impacto medio-intermedio (segundo cuartil año 2018). Se contempla una tercera búsqueda abierta de artículos del ámbito médico indexadas en PubMed para detectar y tratar el posible sesgo derivado.
Resultados y discusión: La MS cardíaca es la más estudiada a nivel nacional, con buena tasa de colaboración entre centros y especialidades, pero, sin traducirse en tasas mayores de artículos originales, pero sí en *artículos especiales* y *case-reports*. A nivel internacional los *case-reports* son el formato predominante, seguidos de los *case-reports*, sobretodo centrados en MS no cardíaca.
Conclusiones: Como perspectiva de futuro, sugerimos potenciar las políticas dirigidas a fomentar la investigación y sobretodo a la creación de redes de colaboración con otras especialidades, con el fin de dar un enfoque multidisciplinar preventivo y clínico a la investigación en MS.

Introduction

Legal and forensic medicine is one of the disciplines which are termed medical-social, as the social context is its objective, going beyond the individual. Forensic medicine is relevant firstly because clinical and biological expertise is necessary to resolve certain legal problems that would otherwise be left unresolved. Secondly, there is the theoretical function that medicine brings by supplying biological and medical knowledge for the development and continuous improvement of law.[1]

Legal and forensic medicine in Spain is currently structured around the Legal Medicine and Forensic Science Institutes (IMLCF) and the Nacional Institute of Toxicology and Forensic Sciences (INTCF). These institutes fulfil research and teaching functions, as well as the work of helping judicial bodies.[1–4] This leads to the publication of scientific studies, works and other regular academic activities that are important not only in the purely scientific world of knowledge, but also in epidemiology and public health policies.[5]

Forensic pathology is one of the disciplines that in recent years has undergone the greatest scientific and potential growth. Within this field, one of the subjects covered the most often is sudden death (SD). SD accounts for 12% of natural deaths (300,000 deaths/year in the United States alone), and it causes a strong social and economic impact. As it is a complex cause of death, it unites the clinical, pathological, epidemiological and preventive fields.[6,7] SD has classically been divided into adult and infant SD, while adult SD has been subdivided into cardiac SD (CSD), which accounts for up to 80%-90% of cases, and non-cardiac SD (NCSD).[8] Studies of SD have evolved in their approach, as morphological studies predominated in the past and now a genetic-molecular approach is increasingly being used; these forms of approach are more appropriate and are more important for the clinical and preventive fields (channelopathy and family member studies).[9–11]

The aim of this work is to analyse Spanish forensic scientific production in the field of SD, one of the most paradigmatic areas of legal and forensic medicine, and also one which has undergone the greatest development over recent years. More specifically, it studies scientific production over the last 10 years, comparing it with foreign production over the same period.

It is both necessary and appropriate 15 years after the creation of the IMLCF to examine their scientific aspects and functions.[2,12,13] As far as we know, no studies have analysed forensic scientific publications in Spain; the results may elucidate the state of health of research, as well as detecting current cutting-edge or hot topics where further research is required, with the aim of offering constructive criticism that leads to future improvement.

Material and methods

This study is based on the assumption that the Spanish production of scientific papers on the subject of SD is an indicator of the quality and level of forensic research in our country, and it uses this criterion to compare it with scientific production abroad. Both of the most important Spanish journals in the field of legal and forensic medicine were used for this study: the *Revista Española de Medicina Legal* and the *Cuadernos de Medicina Forense*. Both journals

Table 1 Variables studied.

Year of publication
Type of paper (original, review, series of cases, special paper, others)
Type of sudden death (CSD vs NCSD)
CSD sub-topics
NCSD sub-topics
Population studied (adult, non-adult)
Forensic approach (macroscopic, macroscopic and microscopic, multiple studies)
Study design (single/multicentre, single/multidisciplinary)
Main speciality of the authors (forensic medicine vs clinical medicine)
Number of (sudden death) cases studied

are indexed in SCOPUS, IBECS (the Spanish Health Sciences Bibliographical Index) and the IME (Spanish Medical Index).

The Journal of Forensic Science was selected for the purposes of comparison, as an international publication indexed in the Science Citation Index (SCI) with an intermediate impact factor (quartile 2 in 2018; Web of Science),[14] as it is representative of international journals in general. All of the journals publish a comparable profile of papers.

The search criteria were the key words ''*Muerte súbita*'' / ''Sudden death'', considering a 10 year time period from 2009 to 2018 (both inclusive). The resulting papers were always selected on condition that they contained the key words in their title, abstract or key words.

Table 1 shows the set of variables studied in each one of the papers in both groups. Comparative statistical analysis was undertaken using Pearson's chi-squared test and the SPSS package.

To detect the risk of distortion in selection arising from have restricted the search for SD to solely two Spanish forensic journals that are not SCI-indexed, PubMed was searched for papers in indexed medical journals using the descriptors ''Sudden Death'', ''Spain'' and ''Forensic Science'', considering the 10-year time period (2009–2018). The crosses used were: ''Sudden Death AND Spain'' and ''Sudden Death AND Forensic Science AND Spain''. The resulting papers were always selected on condition that they included the key words ''Sudden Death'' in their title, abstract or key words.

Results

A total of 99 papers were analysed in the Journal of Forensic Science (international group) and 49 papers in the Spanish group: *Revista Española de Medicina Legal* (n = 34) and *Cuadernos de Medicina Forense* (n = 15). A total of 1537 instances of studied SD were found in all of the papers in both groups. Table 2 shows the summary of papers according to variable and group of journals.

The results show that in Spain the greater part of scientific production on SD is basically concentrated in two autonomous communities: Catalonia (28.9%) and Andalusia (26.7%) (Fig. 1).

In how papers are distributed according to year of publication the irregularity of papers on SD in forensic medicine stands out (Fig. 2). Compared with the international group, the tendency is Spain can be seen to be more regular than it is in other countries. Nevertheless, the differences between both groups are not statistically significant ($P = .117$).

In general there is an outstanding tendency to publish case-reports (26.5% vs 66.7%). In Spain there are more reviews (24.5% vs 4.0%) and technical papers (30.6% vs 2.0%) compared to what occurs in the international group, where there is a greater tendency to publish original papers (25.3% vs 8.2%) (Fig. 3). The differences found for this variable are statistically significant ($P \sim .001$).

The differences are also statistically significant for ''Type of sudden death'' ($P = .04$), with an international tendency for NCSD (66,7%) and a national tendency for CSD (57.1%). However, in ''SD sub-themes'' there are no significant differences between both groups for CSD or NCSD ($P = .234$ and $P = .232$, respectively). The only outstanding tendencies are towards papers on the genetic-structural area in the Spanish group for CSD (41.4%) and a predominance of studies on SD in connection with neoplasms in both groups (27.3% vs 36.8%).

For the ''Population'' variable the 18.4% of Spanish papers that do not specify the population group covered by the study stands out. Nevertheless, in both groups there is a predominance of adult SD (81.6% and 71.4%).

In ''Forensic approach'' a greater tendency towards complex studies stands out in the Spanish papers, accounting for up to 51.2% of papers, although the differences between both groups are not significant here ($P = .367$).

At an international level there is a predominance of publications with a single centre design (77.8% vs 26.5%) and single discipline (72.7% vs 49.0%). This is exactly the opposite of what occurs in Spanish publications, with more multicentre and multidisciplinary studies. This difference is statistically significant ($P = .001$ and $P = .004$, respectively).

Finally, there are no statistically significant differences in the variable ''Chief speciality'' between both groups ($P = .853$), and the sole outstanding fact here is that 16% of the authors are from the clinical rather than the forensic field.

The search for distortion involved searches for crossed descriptors Sudden Death, Spain and Forensic Sciences, giving rise to a total of 223 papers. The outstanding findings of this search were 122 original papers (54.7%), 60 reviews (26.9%), 19 series of cases (8.5%), 1 special paper (0.5%) and 21 ''others'' (9.4%). In this search 93.3% of the main authors work in the clinical area (hospitals or in associated research centres); on the contrary, 6.7% are authors who work in the forensic field (IMLCF and INTCF).

Discussion

This study aims to examine the health of Spanish forensic research by comparing its quality with that of international research. For this it takes the scientific publications on SD as the quality marker, as study of this is becoming increasingly important, especially in terms of epidemiology and prevention. The studies of SD published at a national level in the *Revista Española de Medicina Legal* and in *Cuadernos de Medicina Forense* (the chief forensic reference journals in Spain) were compared with those published internationally, taking the *Journal of Forensic Science* as the reference.

Table 2 Number of papers per variable and group.

	International		Spanish		
	n = 99		n = 49		p
Type of paper					.001
Original	25	(25.3%)	4	(8.2%)	
Review	4	(4.0%)	12	(24.5%)	
Case-report	66	(66.7%)	13	(26.5%)	
Technical-special	2	(2.0%)	15	(30.6%)	
Others (editorials, letters)	2	(2.0%)	5	(10.2%)	
Type of sudden death					.04
Cardiac	32	(32.3%)	28	(57.1%)	.234
Structural-genetic	9	(28.1%)	12	(41.4%)	
Sport-associated	2	(6.3%)	6	(20.7%)	
Multisystem	7	(21.9%)	3	(10.3%)	
Toxic-infectious	4	(12.5%)	2	(6.9%)	
Others (pregnancy...)	10	(31.3%)	6	(20.7%)	
Non-cardiac	66	(66.7%)	19	(38.8%)	.232
Neurological	1	(1.5%)	3	(15.8%)	
Neoplasm	18	(27.3%)	7	(36.8%)	
Metabolic	4	(6.1%)	1	(5.3%)	
Genetic	10	(15.2%)	1	(5.3%)	
Rheumatological	2	(3.0%)	1	(5.3%)	
Vascular	8	(12.1%)	1	(5.3%)	
Toxic-infectious	20	(30.3%)	4	(21.1%)	
Others (digestive)	3	(4.5%)	1	(5.3%)	
Not classifiable	1	(1.0%)	2	(4.1%)	
Population					
Adult	81	(81.6%)	35	(71.4%)	
Non-adult (paediatric, neonatology)	16	(16.2%)	5	(10.2%)	
Not classifiable	2	(2.0%)	9	(18.4%)	
Study design					
Single centre	77	(77.8%)	13	(26.5%)	.001
Multicentre	22	(22.2%)	36	(73.5%)	.001
Single discipline	72	(72.7%)	24	(49.0%)	.004
Multidiscipline	27	(27.3%)	25	(51.0%)	.004
Principle speciality					.85
Forensic medicine	84	(84.8%)	41	(83.7%)	
Clinical/pathological medicine	15	(15.2%)	8	(16.3%)	
Forensic approach[a]	93		43		.37
Macroscopic only	12	(12.9%)	3	(7.0%)	
Macroscopic and microscopic	44	(47.3%)	18	(41.9%)	
Multiple studies[b]	37	(39.8%)	22	(51.2%)	
Cases studied[c]	1.408		129		

[a] Only the papers that contain a forensic study.
[b] Complex forensic study ($\geq$ 1 of the following approaches: radiological, genetic, toxicological, others).
[c] Counting all of the cases in the analysed papers.

The results obtained show differences that help to set clear objectives for the future regarding the tendency or field which research should concentrate on and cover.

The results for autonomous communities show that Catalonia (28.9%) and Andalusia (26.7%) lead scientific production here within Spain. This result may be linked to the fact that the Catalonian IMLCF is the largest and most productive in Spain.[13] On the other hand, it should also be taken into account that the Spanish journals analysed have links to certain communities, so that they may appeal more strongly to local authors. As a whole the production of papers was found to be irregular and discontinuous in all of the autonomous communities, and this phenomenon was also found at an international level, with minimum differences.

Statistically significant particularities were detected respecting the typology of the papers. Thus in Spain special papers predominated, such as case-reports and reviews,

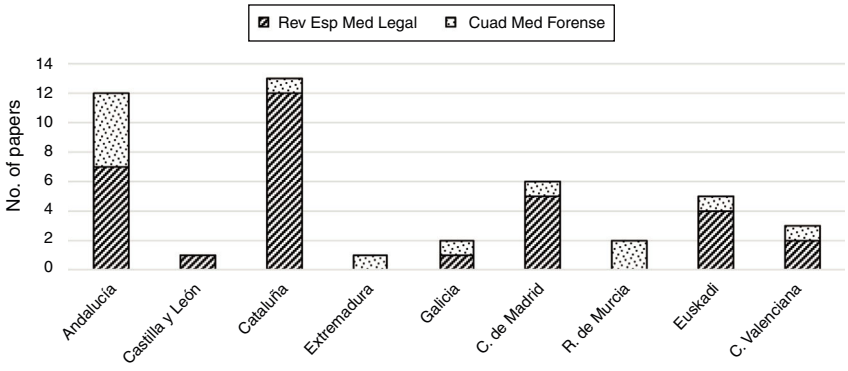

Figure 1 Distribution of papers according to autonomous community.

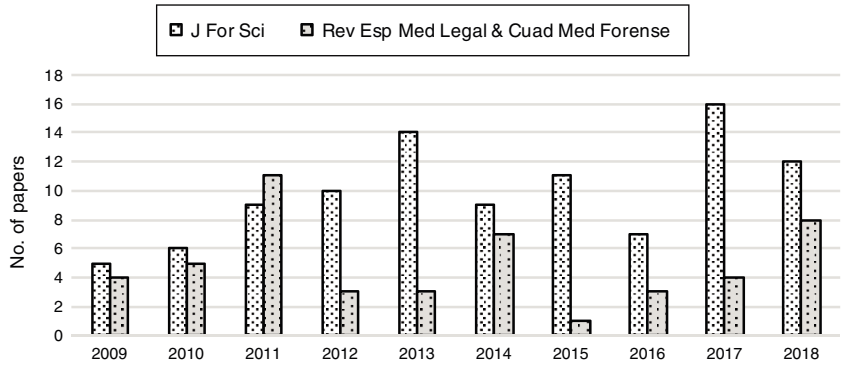

Figure 2 Distribution of papers according to year and group.

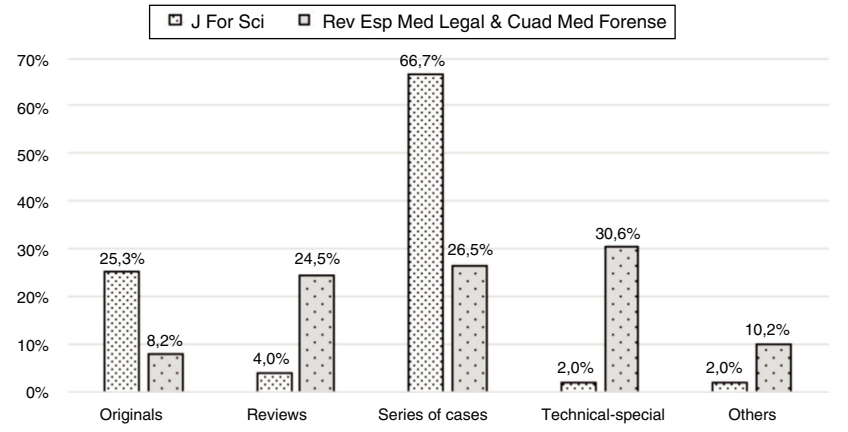

Figure 3 Frequency of types of papers according to group.

followed by original papers. This is an important difference, given that it suggests that even though there is a general predominance of case-reports, the tendency to publish papers that are more robust in scientific terms (prospective and in other original formats) fundamentally applies to the international scene.

It is a widespread practice for forensic science research to be based on knowledge obtained from case-reports, which is logically the preferred format. This format is preferred not only for reasons of convenience, as structural reasons too are important. This is because many items cannot be simulated for evident ethical reasons; case-reports play the role above all of increasing experience-based knowledge.[15] However, their anecdotal nature means they form a basis for solely theoretical construction.[16] Although case-reports make an essential contribution to the speciality, they are the lowest-ranking step in the hierarchy of evidence.[17] As a result of this quality aspects must be taken into consideration and, as Madea recommends,[15] they should form the basis for a systematic study or one based on a hypothesis.

The results show that the predominant type of SD described in Spanish publications is cardiac. This is the opposite of what happens at an international level, where NCSD predominates. This fact reflects the long tradition of

research into CSD which the Spanish state has undertaken for years, with a solid research network, above all in the field of CSD genetics and prevention.[11,18,19] The result of this development is that research into SD is led by the clinical field and is promoted by groups with more scientific resources, above all those deriving from competitive research projects, which leads to greater scientific production.

Regarding the role of the IMLCF and INTCF in these projects and research networks, they have clearly participated in an outstanding way in projects linked to CSD. One example is the research work and long tradition in this field of the Basque Country IMLCF,[20,21] the *Estudio Pheidippides* in the Community of Murcia with the IMLCFM,[22] the *Projecte MOSCAT* in Catalonia with the IMLCFC[23] and the *MUSIB* program of the IMLCF in the Balearic Islands,[24] among others.[25] Nevertheless, these actions seem to be insufficient to place the IMLCF in a leading position in cutting-edge research.

If we consider original papers to be the result of more precise and robust studies, unlike review papers or case-reports, and if we analyse their origin (a clinical context or within the IMLCF), we find that in Spain original papers on CSD are mainly led by clinical groups. When we compare national and international production, the results of our study show that at an international level there is a significantly different tendency to use an original paper format, while at national level we are still producing relatively few such documents. This affects the quality of scientific production unfavourably and raises new challenges for improvement in the future.

Nevertheless, a positive datum within the results obtained shows that in our country there is a greater tendency towards cooperation between centres than is the case internationally. It is significant that there are multicentre and multidisciplinary networks which centre on the study of SD, fundamentally cardiac SD. In the same way, the rates of collaboration between specialities (the clinical and forensic fields) for conjoint publication at a national level are acceptable, and higher than is the case internationally. These results are positive and encouraging, although in the specific case of the Spanish publications, these are not necessarily based on more hypothesis-based or prospective studies, but rather reflect the conjoint publication of papers based on reviews or case-reports. Researchers should strengthen these scientific alliances, delocalise research project leadership and promote a higher profile for forensic science.[9]

There is also clear collaboration between forensic and clinical medicine, as can be seen when the chief speciality of the authors is analysed. A reflection of this collaboration is thus the fact that in 16% of national and international medical-legal or forensic papers the main speciality of the authors is clinical medicine. This co-working between forensic and clinical medicine is the reason why many authors from both fields underline the importance of coordination with public bodies (The Ministries of Justice and Health) and the creation of a national registry of cases, to meet the challenge of SD.[9,26]

The chief limitation of this work is the fact that it centres analysis on two Spanish journals with quality indexes that differ from those of the SCI. The fact that we restrict ourselves to these two journals to reflect and represent Spanish forensic research may form a part of selection bias. As they are journals with less demanding quality indexes it is probable that more robust studies are published by others which are SCI indexed. To correct this distortion we undertook a search for the descriptors Sudden Death, Spain and Forensic Science, considering the last 10 years. A total of 223 papers were found, four times more papers than was the case in the group of national journals. More than half of the papers were original, with a far lower proportion of case-reports. Nevertheless, the majority of the lead authors work in the clinical field, while a minority (6.7%) work in the forensic area. The conclusion is therefore than there is no significant escape of papers with a forensic origin into this ''bias group''. We therefore checked that national Spanish forensic scientific production is mainly published in Spanish journals.

This work was based on the assumption that papers published in the *Revista Española de Medicina Legal* and in *Cuadernos de Medicina Forense* in Spain (even when they are not SCI indexed) are associated with the indexed level of publication of the SCI. We consider that the fact that such a low percentage (6.7%) corresponds to forensic authors suggests that national publications channel a large part of the scientific production which originates directly in the IMLCF. On the other hand, this would also be corroborated by the fact that the author profile we found when searching for bias is different from that of the Spanish group. This suggests that those who publish internationally are not from the forensic field and probably work in the clinical area. Thus this escape or selection bias would not apply to forensic specialists, but rather to clinicians.

It is also acceptable to assume this relationship between what forensic medicine specialists publish at a national level and at an international level. This is probably because the researcher often wishes to publish their work in a medium that is both close and accessible, such as the *Revista Española de Medicina Legal* and *Cuadernos de Medicina Forense*. It should not be forgotten that both journals are very widely read, given their increasing growth not only in Spain but also in Spanish America.

These communications have been returning strongly thanks to the digitisation that has taken place over the past decade.[27,28] According to De Granda[29] and Cabañas et al.[30] it is desirable to continue to work on producing journals that are close and high quality, ones that aspire and eventually achieve an impact and indexing factor in international databases, points that intrinsically indicate quality.

Conclusions

Comparison of current forensic research into SD in Spain and at an international level shows certain significant differences. As a whole, case reports play an important role, as do studies of adult SD and genetic and structural studies of CSD, while there is also more emphasis on more modern and complex forensic methodologies. At a national level there is a greater tendency to publish reviews or special papers, even more so than case-reports, and to publish on CSD. There is also a higher rate of collaboration between centres and specialities (forensic doctors, IMLCF and clinical medicine, hospitals and research centres). At an international level, on the other hand, original studies and those which centre on NCSD above all are more important.

Lastly, when the results as a whole are analysed, we ask ourselves whether the IMLCF are the chief drivers of research in the field of forensic pathology? Research is one of their functions, and they have the necessary resources.

However, we have shown that the standard is set by the clinical field, with occasional co-working and involvement by the IMLCF that should be increased in the future. The role of the IMLCF is often restricted to supplying data corresponding to the victims of SD.

Based on this study we propose a series of recommendations for the future:

1 Clinical professionals should receive good training in legal medicine, to raise their awareness in cases of unexplained or unexpected death.
2 Offer stronger encouragement for the execution of high quality research by legal and forensic medicine professionals.
3 Build up cooperation networks and ensure closer coordination between the judicial and health systems and the other institutions which are involved, to increase data exchange.
4 Spanish journals should continue to work to achieve indexing and obtain a competitive impact factor. The tendency should be not only to encourage research, but also to disseminate the most cutting-edge research and latest hot topics among Spanish forensic professionals.
5 As an expectation for the future, and based on the findings of this study, the interest in genetics may be expected to increase, as will the clinical and preventive application of the results of studies on SD. Forensic medicine professionals have to take part in these processes.
6 Within the field of legal and forensic medicine it is not only necessary to motivate professionals to continue researching, as they should also coordinate better with clinical and care research groups with the aim of creating high quality papers. Within the clinical field it is necessary to continue co-working and discover the links between understanding and specific cases.

Conflict of interests

The authors have no conflict of interests to declare.

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