Notifying patients exposed to blood products associated with Creutzfeldt–Jakob disease: theoretical risk for real people

Evidence

Études

Susan M. King,* MD; Heather Watson,† RN; Helen Heurter,* RN; Maura Ricketts,‡ MD; Susie Elsaadany,‡ MMath

From the Divisions of *Infectious Diseases and of †Haematology, Department of Paediatrics, The Hospital for Sick Children and the University of Toronto, Toronto, Ont., and the ‡Blood-Borne Pathogens Division, Bureau of Infectious Diseases, Laboratory Centre for Disease Control, Health Canada, Ottawa, Ont.

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Abstract

Background: In July 1995 the Canadian Red Cross Society recalled blood products because of the hypothetical risk of transmission of Creutzfeldt–Jakob disease (CJD) through those blood products. The authors undertook a survey to determine the views of patients and parents of patients about being notified that they or their child had received such blood products.

Methods: The study population consisted of 528 transfusion recipients, of whom 453 (85.8%) were under 16 years of age, notified by the Hospital for Sick Children, Toronto, of the CJD recalls in 1995 and 1996. Families attending an information session were asked to complete a self-administered questionnaire (85 cases). Ninety-seven families randomly selected from those who did not attend the session were interviewed by telephone. The questionnaire was adapted from a questionnaire used to evaluate families' responses to notification of transfusion and risk of HIV infection.

Results: More than 80% of the respondents said they wanted to be notified and would want to be notified if there were another recall. On initial receipt of the notification about two-thirds of the respondents had been anxious, fearful or angry. There was no one method of conveying the information that suited all, but a personalized letter was seen as the most acceptable method.

Interpretation: Most parents of children who have received blood products are in favour of being informed about the risk of CJD, despite the uncertainty of the information on risk and the anxiety that such information causes.

Résumé

Contexte : En juillet 1995, la Société canadienne de la Croix-Rouge a rappelé des produits du sang à cause du risque hypothétique de transmission de la maladie de Creutzfeldt–Jakob par les produits du sang en question. Les auteurs ont entrepris un sondage pour déterminer ce que pensent les patients et leurs parents du fait d'avoir été prévenus que leurs enfants ou eux-mêmes avaient reçu de tels produits du sang.

Méthodes : La population à l'étude comportait 528 personnes qui ont reçu une transfusion, dont 453 (85,8 %) avaient moins de 16 ans, que l'Hôpital pour enfants malades à Toronto a prévenues des rappels à cause de la maladie de Creutzfeldt–Jakob survenus en 1995 et en 1996. On a demandé aux familles présentes à une séance d'information de remplir un questionnaire (85 cas). On a interviewé par téléphone 97 familles choisies au hasard parmi celles qui n'avaient pas assisté à la séance d'information. Le questionnaire était tiré d'un questionnaire qui a servi à évaluer les réponses des familles à la notification de transfusion et du risque d'infection par le VIH.

Résultats : Plus de 80 % des répondants ont dit vouloir être prévenus et ont déclaré qu'ils voudraient être prévenus de tout autre rappel. Au sujet de la notification

initiale, environ 66 % des répondants avaient ressenti de l'anxiété, de la crainte ou de la colère. Il n'y avait pas de façon unique de communiquer l'information qui convenait à tous les répondants, mais la lettre personnalisée a été jugée le moyen le plus acceptable.

Interprétation : La plupart des parents d'enfants qui ont reçu des produits du sang souhaitent être informés du risque de transmission de la maladie de Creutzfeldt–Jakob, en dépit de l'incertitude de l'information concernant le risque et de l'anxiété que suscite une telle notification.

In July 1995 the Canadian Red Cross Society recalled some blood products because of the hypothetical risk of transmission of Creutzfeldt–Jakob disease (CJD) through those blood products.[1] At the Hospital for Sick Children, Toronto, we struggled with the issue of notifying recipients of the products.

We reviewed the medical literature for information on CJD, the risk of transmission of the disease through blood products, the methods of diagnosis of the disease and its treatment.[2–13] There were no studies evaluating the outcome of notification in such circumstances, and no Canadian policy regarding notification had been established.[14]

The main reasons for notifying patients about blood products associated with CJD are: 1) patients have a right to know information relevant to their health and 2) providing information (good, bad or uncertain news) is essential for trust between patients and health care providers. The main reasons for not notifying patients are: 1) such information causes anxiety, 2) it is difficult to convey to patients information that has a high level of uncertainty, 3) there is no evidence of any harm due to CJD from these products and 4) since there is no test for CJD, transmission or lack of transmission cannot be determined. Opinions within our institution were almost evenly divided between those for and those against notification, but almost all the parents involved in a preliminary survey were in favour of notification. Therefore, we decided to proceed with notification of recipients.

In view of the lack of evidence of benefit from notification and the divided opinions, we wanted to evaluate the patients' and families' responses to being notified about the recall, so we undertook a survey of those who were notified.

Methods

Population

The survey population consisted of patients or, in the case of patients under 16 years of age, their parents who had been notified that they or their child had received a blood product at the Hospital for Sick Children that had been recalled because of the potential risk of transmission of CJD through that blood product. These recalls occurred between July 1995 and February 1996.

Questionnaire development

The questionnaire was adapted from one used to evaluate families' responses to notification of transfusion and risk of HIV infection.[15] We solicited comments on the modified questionnaire from 7 health care workers and 2 parents whose children had received blood products, and made revisions accordingly. The questionnaire was designed for both self-administration and administration during an interview.

Questionnaire administration

The families who had been notified were invited to attend an information session. Attendees at these sessions were asked whether one family member would complete the self-administered questionnaire (group 1). An equal number of families, selected at random from those who did not attend an information session, were contacted by telephone and interviewed (group 2). The interviewers were 2 nurses with experience in providing information about transfusion.

Statistical analysis

Descriptive analysis of the major variables was performed. We used χ^2 tests, with weighting, to test the differences between the responses of the 2 groups.

Results

Population

Of the 616 recipients of blood products associated with CJD, 281 (45.6%) had received intravenous immunoglobulin therapy, 262 (42.5%) had received albumin, and 73 (11.8%) had received factor VIII concentrate. Patients were excluded from notification if they had died or were receiving palliative care. Of the 528 recipients notified, 75 were 16 years of age or older. Representatives of 100 families attended the information session, 93 of whom agreed to complete the self-administered questionnaire; of these questionnaires, only 85 included enough information to allow analysis (group 1). Of the 100 fami-

lies contacted by telephone, 97 completed the interview (group 2); in the 3 remaining cases the interview was not completed because of language difficulties. Of the overall group of 190 respondents, 12 (6.3%) were 16 years of age or older. The mean interval between notification and response to the questionnaire was 4 weeks for group 1 and 14 weeks for group 2.

Questionnaire responses

The 2 groups did not differ significantly in their responses to the questions concerning knowledge about transfusion and risks, or to those concerning the way they had been notified. Seventy-eight percent (weighted value) of respondents were aware of the transfusion. The most favoured notification method was letter (40%), and the least favoured method was telephone (9%). More than 50% of respondents indicated that their "current" physician (either a community physician or their hospital specialist) should be the person to give them the information or should be involved with the notification process.

Negative emotional responses, reported by about two-thirds of respondents, were more frequent in group 1 than in group 2. In group 2, 44% indicated that their anxiety, fear and anger had decreased in the interval between notification and the interview. Eighty-one percent of families said they wanted to be notified and 84% that they would want to be notified if there were another recall.

Interpretation

To our knowledge this is the first report of patients' and families' responses to being notified of the receipt of blood products recalled because of risk of CJD. In a recent article on the policy of notifying recipients of blood products associated with CJD, Caulfield and colleagues[16] argued that individual notification is not justified. As discussed in their article, several factors need to be weighed to make this difficult decision. Two of the key factors to be considered are what the "reasonable person" would want to know and the benefit versus harm of notification. Although our study has limitations, it does provide information on these 2 factors.

An important limitation of our study is that all the transfusion recipients were children, so the findings may not be generalizable to transfusion recipients of all ages. The number of recipients aged 16 years or older who responded for themselves was too small to be analysed. The responses of parents surveyed before the study and in both groups in the study were consistently in favour of notification. It would be useful to know whether the responses of families who had not been notified would be the same. However, our results suggest that the "reasonable person" in Ontario whose child receives a transfusion would want to be notified of the child's exposure to blood products that had been recalled because of CJD.

Clearly there are people who wish not to be notified of this type of exposure.[17] In our study 9% of respondents indicated that they would not want to be informed of another recall of blood products. This ratio of information seekers to nonseekers is similar to that found in other situations, such as genetic testing for a breast cancer gene.[18] No single process will meet every individual's choice. In a general notification program, for example, some people who want to know will not be informed, and some who do not want to know will be reached. There is also the potential for creating anxiety unnecessarily in a group not exposed to the recalled products.

It has been argued that the anxiety generated by this information may be a harm that outweighs the benefit of providing the information.[16] To address the issue of benefit versus harm, we asked families about their emotional responses when they received the notification. Most of the parents wanted all the information about their child even if that information caused anxiety. This finding is consistent with the Canadian experience of contacting pediatric transfusion recipients to provide information on the risks of transmission of HIV and hepatitis C virus.[14,19] A similar result was also found in a study on the attitude to testing for the breast cancer gene, in which 95% of the subjects were aware of the anxiety that would be caused if the test outcome were positive, but 88% still wanted to undergo testing.[18]

Surveys have shown that the way "bad news" is given to patients is important.[20,21] The literature on understanding patients' decisions indicates that this is an area where measurement of variables is difficult, and perhaps for this reason it has received less attention in medical research.[22,23] Not surprisingly, there is no one way to give such news that is right for all. However, some common themes emerged through the questionnaire responses and the discussions at the information sessions:

- Minimize the delay in providing the information.
- Personalize the way in which the information is given.
- Provide information at a visit to the physician if possible, but if this means a long delay, a letter should be sent before the visit.
- Provide access to further information and counselling, particularly to deal with anxiety.
- Ensure that the treating physician is aware of the notification.

Although our findings support notification for pediatric recipients of blood products associated with CJD, there are still difficult issues to resolve. First, there is a group who do not wish to know, and in a generalized notification such people would be notified. Second, the fac-

tors that determine benefit versus harm depend on the way in which the notification is conducted and the ability of those providing the information to be aware of the emotional consequences for those notified. Third, since notification requires resources and since resources are limited, we must decide on the value of this in relation to other health care expenditures.

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References

1. Creutzfeldt–Jakob disease in Canada. *Can Commun Dis Rep* 1996;22:57-61.
2. Bernoulli C, Siegfried J, Baumgartner G, Regli F, Rabinowicz T, Gajdusek DC, et al. Danger of accidental person-to-person transmission of Creutzfeldt–Jakob disease by surgery. *Lancet* 1977;1:478-9.
3. Brown P, Gibbs CJ, Rodgers-Johnson P, Asher DM, Sulima MP, Bacote A, et al. Human spongiform encephalopathy: the National Institutes of Health series of 300 cases of experimentally transmitted disease. *Ann Neurol* 1994;3:513-5.
4. Brown P, Preece MA, Will RG. "Friendly fire" in medicine: hormones, homograft and Creutzfeldt–Jakob disease. *Lancet* 1992;340:24-7.
5. Rapidly progressive dementia in a patient who received a cadaveric dura mater graft. *MMWR* 1987;36:49-50,55.
6. Cochius JJ, Hyman N, Esiri MM. Creutzfeldt–Jakob disease in a recipient of human pituitary-derived gonadotropin, a second case. *J Neurol Neurosurg Psychiatry* 1992;55:1094-5.
7. Creange A, Gray F, Cesaro P, Adle-Biassette H, Duvoux C, Cherqui D, et al. Creutzfeldt–Jakob disease after liver transplantation. *Ann Neurol* 1995;38:269-72.
8. Duffy P, Wolf J, Collins G, DeVoe AG, Streeten B, Cowen D. Possible person-to-person transmission of Creutzfeldt–Jakob disease [letter]. *N Engl J Med* 1974;290:692.
9. Esmonde TFG, Will RG, Slattery JM, Knight R, Harries-Jones R, de Silva R, et al. Creutzfeldt–Jakob disease and blood transfusion. *Lancet* 1993;341:205-7.
10. Heye N, Hensen S, Muller N. Creutzfeldt–Jakob disease and blood transfusion [letter]. *Lancet* 1994;343:298-9.
11. Manuelides L. The dimensions of Creutzfeldt–Jakob disease. *Transfusion* 1994;34:915-28.
12. Tange RA, Troost D, Limburg M. Progressive fatal dementia (Creutzfeldt–Jakob disease) in a patient who received homograft tissue for tympanic membrane closure. *Eur Arch Otorhinolaryngol* 1990;247:199-201.
13. Will RG. Epidemiology of Creutzfeldt–Jakob disease. *Br Med Bull* 1993; 49:960-70.
14. Laboratory Centre for Disease Control, Health Canada. *Summary — consensus conference. Creutzfeldt–Jakob disease — decision-making in times of uncertainty, Toronto, June 5-6, 1996.* Ottawa: Health Canada; 1996.
15. King SM, Murphy T, Corey M, Newman AM, Major C, McCrindle BW, et al. The HIV Information Project for transfusion recipients a decade after the transfusion. *Arch Pediatr Adolesc Med* 1995;149:680-5.
16. Caulfield T, Dossetor J, Boshkov L, Hannon J, Sawyer D, Robertson G. Notifying patients exposed to blood products associated with Creutzfeldt–Jakob disease: integrating science, legal duties and ethical mandates. *CMAJ* 1997;157:1389-92.
17. Walker R. Man's life shaken by blood danger warning: he opposes lawsuit that would force disclosure. *Calgary Herald* 1996 Feb 9;Sect A:3.
18. Julian-Reynier C, Eisenger F, Vennin P, Chabal F, Aurran Y, Nogues C, et al. Attitudes towards cancer predictive testing and transmission of information to the family. *J Med Genet* 1996;33:731-6.
19. Heddle N, Kelton JG, Smaill F, Foss K, Everson J, Janzen C, et al. A Canadian hospital-based HIV/hepatitis C look-back notification program. *CMAJ* 1997;157:149-54.
20. Krahn GL, Hallum A, Kime C. Are there good ways to give "bad news"? *Pediatrics* 1993;91:578-82.
21. Strauss RP, Sharp MC, Lorch SC, Kachalia B. Physicians and the communication of "bad news": parent experiences of being informed of their child's cleft lip and/or palate. *Pediatrics* 1995;96:82-9.
22. Redelmeier DA, Rozin P, Kahneman D. Understanding patients' decisions. Cognitive and emotional perspectives. *JAMA* 1993;270:72-6.
23. Health Services Research Group. Studying patients' preferences in health care decision making. *CMAJ* 1992;147(6):859-64.

Reprint requests to: *Dr. Susan M. King, Division of Infectious Diseases, The Hospital for Sick Children, 555 University Ave., Toronto ON M5G 1X8; fax 416 813-5032*