Response Bias in the Nationwide Epidemiological Survey of an Intractable Disease in Japan

Shuji Hashimoto1, Kazuo Fukutomi2, Masaki Nagai3, Yasukazu Nakamura3, Hiroshi Yanagawa3, Ryutiro Sasaki4, Yoshiyuki Ohno4, and Kunio Aoki5

Annual numbers of patients with the so-called intractable diseases were estimated in the nationwide epidemiological surveys under the assumption that the mean number of patients among "responded" hospitals is equal to that among "non-responded" hospitals. This paper evaluated the validity of this assumption, using the data of idiopathic thrombocytic purpura (ITP) obtained from the nationwide epidemiological survey and the number of ITP patients who are financially subsidized for treatment (FST-patients). The mean numbers of ITP FST-patients among "responded" hospitals in the nationwide survey were compared with the mean numbers among "non-responded" hospitals. The ratio of the mean number among "responded" hospitals to that among all hospitals was found to be between 1.0 and 1.1, suggesting that this assumption was sufficiently valid. J Epidemiol, 1991; 1: 27-30.

response bias, estimation, number of patients, intractable diseases, nationwide survey

Since 1972, the Ministry of Health and Welfare of Japan has promoted researches in the causes and controls of the so-called intractable diseases of unknown etiology and of no specific medical treatment. To investigate the epidemiological features of intractable diseases (prevalence, age distribution, sex ratio, etc.), the Research Committee on Epidemiology of Intractable Diseases, in cooperation with the Research Committees on each specific intractable diseases, has conducted several epidemiological surveys1-3. The method used in these surveys was to send a postal questionnaire to all hospitals with 200 beds or more throughout Japan, where most patients are actually treated1,4, inquiring for the numbers of patients treated during the preceding year. The response rates have usually been 50-60% in these surveys1-3,5.

The data obtained from the surveys provided the opportunity of estimating the prevalence of intractable diseases under the assumption that the mean number of patients among "responded" hospitals to the survey is equal to that among "non-responded" hospitals. Although the prevalence of intractable diseases has been estimated under this assumption1-3, the validity of this assumption has not been evaluated4. To confirm the validity of this assumption, it would be necessary to find the numbers of patients with intractable diseases in all hospitals and clinics, including "non-responded" hospitals. Patients with some intractable diseases specified by the Ministry of Health and Welfare of Japan can partially subsidized for the medical care costs which are not covered by health insurance. Although the number of patients who are financially subsidized for treatment (denoted as "FST-patients") is not the total number of all patients, the number of FST-patients in each hospital and clinics was obtainable by the list of all patients registered in all prefectural governments throughout Japan6,7.

This paper evaluated the validity of "the assumption of no response bias" in the nationwide epidemiological surveys of intractable diseases. Using the data of idiopathic thrombocytopenic purpura (ITP) obtained from the nationwide epidemiological survey7 as an
example and the number of ITP FST-patients, the mean number of FST-patients with ITP among "responded" hospitals to the nationwide survey was compared with the mean number among "non-responded" hospitals. If the mean number of FST-patients among "responded" hospitals was approximately the same as that in "non-responded" hospitals, it could be concluded that the assumption was correct and the method of estimating prevalence under this assumption was valid.

**MATERIALS AND METHODS**

The data used in this study consisted of information obtained, by "responded" and "non-responded" hospitals, from the nationwide epidemiological survey on ITP conducted in November 1984 and the number of FST-patients with ITP in each hospital in 1984. Among hospitals with 200 beds or more, those in Miyagi prefecture were excluded from our study, since the survey method used in this prefecture differed from that used in other prefectures. The number of hospitals included in our study totaled 1696, excluding 39 in Miyagi prefecture.

The response rates and the numbers of ITP patients reported in the nationwide survey are shown in Table 1. The overall response rate was 57.6%, and the total number of ITP patients reported was 3859. Based on the assumption of no response bias, the number of ITP patients was estimated by dividing the number reported by the response rates in the departments of internal medicine and pediatrics in two types (general and university) of hospitals. The total number of ITP patients was estimated as 6700.

The mean numbers of FST-patients with ITP were calculated for each of "responded" hospitals, "non-responded" hospitals, and all hospitals. The ratios of the mean number of FST-patients among "responded" hospitals and "non-responded" hospitals to the mean number of FST-patients in all hospitals were calculated, because if these ratios are equal to 1.0, the assumption of no response bias would be concluded to be correct.

We obtained the information on the response to the nationwide survey only from the departments of internal medicine and pediatrics in each hospital, but the department in which most FST-patients were treated are actually unknown. Hospitals were, therefore, stratified as those with the departments of internal medicine and pediatrics as well and those with only one or the other. Furthermore, hospitals with multiple departments of internal medicine (this would be found in university hospitals), were stratified as those with 1-2 or 3-6 departments of internal medicine. In each stratum, hospitals were classified as "responded" or "non-responded" by the departments of internal medicine and/or pediatrics. The hospitals with multiple departments of internal medicine, were classified as those with the response rate of more than 50% or those with the response rate of less than 50%.

**RESULTS**

Numbers of departments, numbers of FST-patients, mean numbers of FST patients, and their ratios to the mean number of FST-patients among all hospitals in each stratum are summarized in Table 2.

In the stratum of general hospitals with departments of internal medicine and pediatrics as well, for example, the ratio of mean number of FST-patients was found to be 1.03 when both departments responded, 1.04 when only departments of internal medicine did, 0.94 when only departments of pediatrics did, and 0.93 when both departments did not. In each stratum of hospitals, when excluding the university hospitals with 1-2 departments of internal medicine and with a department of pediatrics, the mean numbers of FST-patients were larger in hospitals with a higher response rate than in those with a lower response rate.

The number of FST-patients was 4251 in general hospitals with department of internal medicine and department of pediatrics, and 1862 in the university hospitals with 3-6 departments of internal medicine and department of pediatrics. The proportion of

| Table 1. Response rates, numbers reported and estimated numbers of ITP patients. |
|----------------------------------|-----------------|-----------------|-----------------|-----------------|-----------------|-----------------|
|                                  | General hospitals | University hospitals |
|                                  | Departments of internal medicine | Departments of pediatrics | Departments of internal medicine | Departments of pediatrics | Total |
| No. of departments               | 1,590            | 1,091            | 283             | 93              | 3,057           |
| No. of "responded" departments   | 972              | 382              | 141             | 66              | 1,761           |
| Response rate (%)                | 61.1             | 33.3             | 49.8            | 71.0            | 57.6            |
| No. of patients reported         | 1,755            | 970              | 774             | 360             | 3,859           |
| Estimated no. of patients a      | 2,900            | 1,800            | 1,600           | 900             | 6,700           |

a Estimated no. of patients = (No. of patients reported)/(Response rate)
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Table 2. Mean numbers of ITP FST-patients and their ratios.

| Strata                          | Responded or not responded to the nationwide survey on ITP | No. of departments | No. of FST-patients | Mean no. of FST-patients | Ratio* |
|---------------------------------|----------------------------------------------------------|--------------------|---------------------|--------------------------|--------|
| General hospitals               |                                                          |                    |                     |                          |        |
| with departments of internal medicine and pediatrics | responded | responded | 718 | 1,473 | 2.05 | 1.05 |
|                                 | responded | not responded | 604 | 1,228 | 2.03 | 1.04 |
|                                 | not responded | responded | 440 | 803 | 1.83 | 0.93 |
|                                 | not responded | not responded | 412 | 747 | 1.81 | 0.93 |
|                                 | total |                                                            | 2,174 | 4,258 | 1.96 | 1 |
| General hospitals               | responded |                                                            | 311 | 114 | 0.37 | 1.07 |
| with a department of internal medicine | not responded |                                                            | 192 | 59 | 0.31 | 0.89 |
|                                 | total |                                                            | 503 | 173 | 0.34 | 1 |
| General hospitals               | responded |                                                            | 3 | 31 | 10.33 | 1.33 |
| with a department of pediatrics  | not responded |                                                            | 1 | 0 | 0.00 | 0.00 |
|                                 | total |                                                            | 4 | 31 | 7.75 | 1 |
| University hospitals            | more than 50% responded |                                                            | 103 | 599 | 5.82 | 1.02 |
| with 3-6 departments of internal medicine and a department of pediatrics | more than 50% not responded |                                                            | 54 | 329 | 6.09 | 1.07 |
|                                 | less than 50% responded |                                                            | 115 | 383 | 3.07 | 0.89 |
|                                 | less than 50% not responded |                                                            | 54 | 351 | 6.50 | 1.14 |
|                                 | total |                                                            | 326 | 1,862 | 5.71 | 1 |
| University hospitals            | more than 50% responded |                                                            | 26 | 92 | 3.54 | 0.92 |
| with 1-2 departments of internal medicine and a department of pediatrics | more than 50% not responded |                                                            | 3 | 9 | 3.00 | 0.78 |
|                                 | less than 50% responded |                                                            | 12 | 57 | 4.75 | 1.23 |
|                                 | less than 50% not responded |                                                            | 0 | — | — | — |
|                                 | total |                                                            | 41 | 158 | 3.85 | 1 |
| University hospitals            | responded |                                                            | 2 | 7 | 3.50 | 2.10 |
| with a department of internal medicine | not responded |                                                            | 7 | 8 | -1.14 | 0.69 |
|                                 | total |                                                            | 9 | 15 | 1.67 | 1 |

The patients who are financially subsidized for treatment were denoted as "FST-patients".
*Ratio of mean no. of FST-patients to the mean number in all hospitals in each stratum
*Response rate

FST-patients in these two strata among all FST-patients was accounted for more than 90%. In these two strata, the ratios of mean number in hospitals with a higher response rate were between 1.0 and 1.1.

DISCUSSION

In the present study, mean numbers of ITP FST-patients were shown to be larger in hospitals with a higher response rate than in hospitals with a lower response rate. This suggests that the number of ITP patients was overestimated under the assumption of no response bias. When the number of patients is estimated by dividing the number reported by the response rate, the magnitude of overestimation is equal to the ratio of the mean number of patients among "responded" hospitals to that among all hospitals.

The ratio of mean number of ITP FST-patients among "responded" hospitals to that among all hospitals was found to be between 1.0 and 1.1 in the majority of strata for ITP FST-patients. The number of FST-patients was fewer than the total number of all patients with ITP, but should account for a large proportion of the total, since health insurance for patients of less than 70 years of age does not fully cover medical costs and the proportion of patients of less than 70 years of age among all patients with ITP was reported to be more than 90%. Response or non-response by hospitals to the nationwide survey does not depend on whether or not the medical expenses of patients treated are subsidized i.e., whether or not they are FST-patients. This suggests that the estimated number of ITP patients is about 0-10% greater than the true number. The method of estimating the number...
of ITP patients under the assumption of no response bias is believed to be almost completely valid.

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