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Letter to the Editor

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Underestimation of Trisomy 18 and 13 Syndromes in Vital Statistics from Inadequate Death Certificates

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Dear Editor,

We read the article entitled "Population-based analysis of secular trends in age at death in trisomy 18 syndrome in Japan from 1975 to 2016" with great interest [1]. The article suggested that the survival time of infants with trisomy 18 syndrome (T18) has been increasing. Furthermore, with the death certificate data linked to the vital statistics database for 2014–2016, the authors included fatalities with T18 listed as a contributory cause, not the primary cause. They concluded that patients with T18 assigned as a contributory cause of death had significantly longer median survival time than those with T18 as the primary cause of death.

Child death review was performed in Shiga Prefecture, Japan, which has a population of approximately 1.4 million people and 12,000 annual births. All death certificates for victims younger than 18 years of age submitted to Shiga Prefecture between 2015 and 2020 were reviewed by the experts of Forensic Medicine and Pediatrics. Among these cases, we chose to examine those that included T18 and trisomy 13 syndrome (T13). Overall, 7 cases with T18 and 5 with T13 were obtained. T18 was listed as the primary cause of death in 4 cases, but was included as a contributory disease in 3 cases. When reviewing these descriptions, these 3 death certificates had not been correctly generated according to the manual [2]. In the first case, although the patient died of an anoxic spell associated with double-outlet right ventricle from T18, T18 was not listed as the primary cause, but rather as a contributory factor. In the other 2 cases, the patients died of upper airway obstruction associated with nasopharyngeal stenosis, or ventricular septal defect from T18. Yet, T18 was not listed as the primary cause. In 3 of 5 fatal cases of T13, T13 was listed as a condition contributing to death, despite the patients dying from dilated cardiomyopathy, tetralogy of Fallot, or double-outlet right ventricle caused by T13 (unpublished data).

According to our population-based study, the significant underestimation of the number of T18 patients included in vital statistics because of inaccurate instructions for the cause of death section of the death certificate has had a substantial impact on the results of the previous analysis [1]. Pediatricians possibly do not include T18 and T13 as the primary cause of death in certain cases because these diseases cause a variety of external and organ malformations. The differences between cases in which T18 or T13 was included as a primary cause or a contributing disease could result from inadequate instruction. The cause of death data includes in death certificates are reportedly full of errors [3, 4]. Knowing the epidemio-

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logical background and changes in the course and outcome of T18 and T13 are valuable for medical interventions and improving the quality of life of patients and their families. To accumulate proper evidence, correct instructions for generating death certificates are absolutely crucial. We suggest that standardized instructions regarding correct descriptions should be promoted.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Kohei Takashima contributed to conceptualization, investigation and analysis, and writing original draft preparation. Masahito Hitosugi contributed to methodology, data curation, writing, review, and editing.

References

1 Suto M, Isayama T, Morisaki N. Population-based analysis of secular trends in age at death in trisomy 18 syndrome in Japan from 1975 to 2016. Neonatology. 2021;118(1):47–53.
2 Department of health and human services, Centers for Disease Control and Prevention, National Center for Health Statistics. Physicians' Handbook on Medical Certification of Death. 2003 Revision.
3 Mieno MN, Tanaka N, Arai T, Kawahara T, Kuchiba A, Ishikawa S, et al. Accuracy of death certificates and assessment of factors for misclassification of underlying cause of death. J Epidemiol. 2016;26(4):191–8.
4 Schuppener LM, Olson K, Brooks EG. Death certification: errors and interventions. Clin Med Res. 2020 Mar;18(1):21–6.