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# Healthcare service use and medical outcomes of tracheostomy-dependent children: a nationwide study

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|                               |   |





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for Review Only

Healthcare service use and medical outcomes of tracheostomy-dependent children: a nationwide study

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15 ABSTRACT

Background: Despite the rising incidence, there is a lack of comprehensive resources for families to navigate the challenges of living with a tracheostomy, emphasising the need for evidencebased support in understanding post-operative care and long-term adjustments. This study aimed to examine the pattern of utilising healthcare services and nationwide medical outcomes in children who underwent a tracheotomy before the age of 2 years.

Methods: This retrospective study used the National Health Insurance System database from 2008 to 2016 and included all children codified with tracheotomy procedure codes before their second birthday. Healthcare utilisation, such as medical costs, the number of hospital visits, home healthcare nursing, and the medical diagnoses upon readmission in the first 2 years after tracheotomy, were evaluated. Multivariable logistic regression analysis was used to determine the factors affecting mortality.

Results: In total, 813 patients were included in this study. Their use of healthcare services
and the accompanying expenses were higher than the national medians for similar age groups;
however, both metrics decreased in the second year. The major causes of admission within 2 years of
surgery were respiratory and neurological diseases. The mortality rate within 2 years was 37.8%.
Higher risks of mortality were associated with having two or more complex chronic conditions. Use
of home healthcare nursing services was associated with a lower mortality risk.

34 Conclusion: Paediatric patients with more complex chronic conditions tended to have higher
35 mortality rates within 2 years after surgery. However, receiving home healthcare nursing was
36 significantly associated with a reduced risk of death. Many causes of hospitalisation may be
37 preventable with education and supportive care. Therefore, further research for establishing an
38 integrated care system for these patients and their caregivers is required.

40 Keywords: Tracheotomy; Paediatrics; Health Services; Home Care Services; Mortality
41

42 Key Messages

| 1<br>2   |    |   |
|--|----|---|
| 2<br>3<br>4  | 43 | What is already known on this topic   |
| 5<br>6   | 44 | Children under the age of 2 who undergo tracheotomy experience high morbidity, significant            |
| 7<br>8   | 45 | complications, and place a substantial burden on their families.                                      |
| 9<br>10  | 46 |   |
| 11<br>12   | 47 | What this study adds  |
| 13<br>14   | 48 | Many causes of hospitalisation may be preventable with education and supportive care. Receiving       |
| 15<br>16   | 49 | home healthcare nursing was significantly associated with a reduced risk of death.                    |
| 17<br>18   | 50 |   |
| 19<br>20<br>21   | 51 | How this study might affect research, practice or policy  |
| 21<br>22<br>23   | 52 | The findings of this study can serve as empirical evidence for establishing new systems, such as home |
| 24<br>25   | 53 | healthcare services, in countries with weak support systems for children who undergo tracheotomy.     |
| 26<br>27<br>28<br>29<br>30<br>31<br>32<br>33<br>34<br>35<br>36<br>37<br>38<br>39<br>40<br>41<br>42<br>43<br>44<br>45<br>46<br>47<br>48<br>49<br>50<br>51<br>52<br>53<br>54<br>55<br>56<br>57<br>58<br>59<br>60 |    | healthcare services, in countries with weak support systems for children who undergo tracheotomy.     |

#### 55 INTRODUCTION

With the development of medical science, the number of medically complex and technology-dependent children has steadily increased. Paediatric tracheostomy is technically difficult, with a higher risk of mortality and perioperative complications than that in the adult population [1, 2]. particularly for younger children. Data from the National Surgical Quality Improvement Program of the American College of Surgeons indicate that children aged <2 years with tracheostomy have higher morbidity, with 24.3% experiencing a major complication within 30 days of tracheotomy [3]. Nonetheless, the procedure is becoming more prevalent in paediatric populations with complex medical conditions or chronic illnesses, such as airway obstruction, cardiopulmonary disease, and neurological impairment [1, 2, 4].

Living with a tracheostomy poses significant challenges for both patients and their families; therefore, they should receive comprehensive information before the procedure. Providing practical insights into the journey after tracheostomy can empower parents to make informed decisions about the surgery and prepare for the required long-term adjustments. It becomes imperative to develop family-centred resources grounded in robust, evidence-based data on contemporary tracheotomy-related healthcare and outcomes. While there is a wealth of literature on paediatric tracheotomy procedures, indications, and complications, studies focusing on the enduring experiences of patients and their families after tracheotomy are lacking [1, 5-7]. This study leveraged the Korean National Health Insurance System (KNHIS) database to examine nationwide healthcare utilisation and outcomes among children who underwent tracheotomy before their second birthday.

**METHODS** 

#### 77 Data source and study design

This retrospective study analysed the data of children born between 2008 and 2016 using a nationwide
database from the Republic of Korea (Korea) and followed their records until 2018. The KNHIS
database collects mandatory information on beneficiaries of national health insurance and medical aid
in a unified manner. This database comprises several linked datasets such as social and economic

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qualifications, medical treatment, results of medical check-ups, and costs [8]. Diagnostic codes are based on the World Health Organization's International Statistical Classification of Disease and Related Health Problems 10th revision (ICD-10). Information on the population census and national annual medical fees was obtained from the Korean Statistical Information Service [9]. The institutional review board of Seoul National University Hospital reviewed and exempted this study from formal approval as the KNHIS database does not contain any identifiable information (No. 2208-153-1353). The study was conducted according to the principles of the Declaration of Helsinki. Children who underwent tracheotomy before the age of 2 were included to focus on a population where tracheotomy complications are more prevalent [3]. We excluded children who died during hospitalisation to ensure that the study assessed progress after elective tracheotomy, which would be more informative in terms of long-term implications. Terminology Tracheotomy cases were defined as those with a corresponding procedural code (O1300, O1301, O1303, O1305, O1306, M5830). Complex chronic conditions (CCCs) are defined as "any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or one organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center" [10]. The list of categories was adopted from CCC version 2 [11]. Patients living with CCCs were defined as individuals assigned CCC disease codes in either the primary or additional diagnosis field for hospital visits. The number of CCCs was defined as the number of CCC categories designated for each patient. Ambulatory care sensitive conditions (ACSCs) refer to clinical conditions wherein the likelihood of an unplanned hospitalisation can be diminished through prompt and efficient outpatient care. Hospitalisation stemming from ACSC may signify a lost opportunity for prevention and an adverse encounter for a child and their family. Additionally, it may signal a deficiency in, or limited access to, high-quality outpatient healthcare services. We defined ACSCs using the patients' ICD-10

109 codes, according to previous studies [12, 13].

110 The relevance index indicates the number of patients in a certain area who used hospitals in 111 the same region. This was calculated by dividing the number of patients who underwent tracheotomy 112 in a hospital located in the same residential area by the total number of patients who underwent 113 tracheotomy residing in that area for the same year. Therefore, a lower relevance index in a specific 114 region indicated that more patients moved to a remote area for treatment [14, 15].

116 Variables

Demographic characteristics included patients' sex, age at tracheotomy, income level, categories and number of CCCs, address of the patient's residence, and the medical institution where tracheotomy was performed. The insurance type (national health insurance or medical aid) and the NHIS premium based on income levels were utilised as proxy indicators for the financial status. The lowest income category was designated as those receiving medical aid. KNHIS income levels were divided into four groups: category 1 (<25% premium); category 2 (25%-50% premium); category 3 (50%-75% premium); and category 4 (>75% premium). Medical aid and category 1 patients were merged for analysis because of the small number receiving medical aid. The country was divided into administrative districts, and patients' residential districts were classified as metropolitan (Seoul, Busan, Incheon, Daegu, Daejeon, Gwangju, Ulsan) or other regions. 

For 2 years following tracheotomy, healthcare utilisation indicators included medical expenses and the combined count of hospitalisations, outpatient visits, and use of emergency medical services. Notably, admissions through the emergency room were recorded under hospitalisations in the database, precluding a separate tally for emergency department visits. Additionally, home nursing care utilisation was identified by assignment of procedure codes (AN200, AN300, AN400, AN500).

# 133 Outcome measures

The main outcome measure was mortality within 2 years of tracheotomy. The primary and top 5
 secondary diagnoses for admission were identified using ICD-10 diagnostic codes. The top 30 codes

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| 54<br>55<br>56 |            |             | Birth (n)     | Tracheotomy<br>(n)                     | Death within<br>tracheotomy<br>episode <sup>*</sup> (n, %) | Death within 2 years<br>after 1 <sup>st</sup> discharge (n, %) |
|----------------|------------|-------------|---------------|--|--|--|
| 51<br>52<br>53 | 159        | Table 1.    | Number of     | births and trache                      | eotomies between 20  | 08 and 2016 in the Republic of Korea                           |
| 49<br>50       | 158        |             |               |  |  |  |
| 47<br>48       | 157        | the 2-yea   | r mortality   | rates (P=0.099).                       |  |  |
| 45<br>46       | 156        | tracheoto   | omy. The ov   | verall mortality ra                    | ate was 37.8%. There                                       | e were no significant annual differences in                    |
| 42<br>43<br>44 | 155        | excluded    | , and 813 w   | vere included. Th                      | ree hundred and sev  | en patients died within 2 years after                          |
| 40<br>41<br>42 | 154        | over the    | years (Figu   | re 1B). Sixty-one                      | e children who died d                                      | during hospitalisation for tracheotomy were                    |
| 38<br>39       | 153        | Table 1).   | However,      | the number and i                       | incidence rate of trac                                     | heotomies in patients <2 years also increased                  |
| 36<br>37       | 152        | In total, 4 | 4,105,326 ir  | nfants were born                       | between 2008 and 2   | 016, with a downward trend (Figure 1A,                         |
| 34<br>35       | 151        | Trends i    | n the numl    | ber of tracheoto                       | mies and mortality   | rates  |
| 31<br>32<br>33 | 150        | RESUL       | ГS            |  |  |  |
| 29<br>30       | 140        | r attents a |               |  | a in this study.   |  |
| 27<br>28       | 147<br>148 |             | -             | <b>involvement</b><br>were not involve | d in this study  |  |
| 25<br>26       | 146        | D (: (      |               |  |  |  |
| 22<br>23<br>24 | 145        | associatio  | ons.          |  |  |  |
| 20<br>21       | 144        | × ×         | ,             | % confidence in                        | tervals (CIs) were ca                                      | lculated to elucidate the strengths of the                     |
| 18<br>19       | 143        |             |               |  |  | onsidered statistically significant, and odds                  |
| 16<br>17       | 142        | outcome     | and predict   | ive factors. All a                     | analyses were perform                                      | ned using SAS software (version 9.4; SAS                       |
| 13<br>14<br>15 | 141        | chi-squar   | re test. Mult | tivariate logistic                     | regression was used  | to evaluate the association between the main                   |
| 11<br>12       | 140        | The annu    | al different  | e in the 2-year n                      | nortality rate after tra                                   | acheotomy was examined using Pearson's                         |
| 9<br>10        | 139        | Statistic   | al analysis   |  |  |  |
| 7<br>8         | 138        |             |               |  |  |  |
| 5<br>6         | 137        | underlyir   | ng medical    | conditions, such                       | as hypoxic ischemic  | encephalopathy.  |
| 2<br>3<br>4    | 136        | were sele   | ected after e | excluding codes r                      | relating to health stat                                    | us/services (tracheostomy status) and                          |
| 1<br>ว         |            |             |               |  |  |  |

5 (7.0)

24 (36.4)

| 2009  | 448,459   | 77  | 10 (13.0) | 26 (38.8)  |
|-------|-----------|-----|-----------|------------|
| 2010  | 474,435   | 87  | 3 (3.4)   | 43 (51.2)  |
| 2011  | 476,710   | 99  | 5 (5.1)   | 29 (30.9)  |
| 2012  | 490,472   | 93  | 3 (3.2)   | 31 (34.4)  |
| 2013  | 442,418   | 117 | 6 (5.1)   | 36 (32.4)  |
| 2014  | 442,341   | 108 | 9 (8.3)   | 36 (36.4)  |
| 2015  | 446,160   | 113 | 8 (7.1)   | 36 (34.3)  |
| 2016  | 415,083   | 109 | 12 (11.0) | 46 (47.4)  |
| Total | 4,105,326 | 874 | 61 (7.0)  | 307 (37.8) |

\* Number of children who died during hospitalisation for tracheotomy and these were excluded in this

**Demographic characteristics** 

study.

Most patients underwent tracheotomy before 1 year of age. The majority (91.4%) had at least one
CCC, and approximately two-thirds had multiple CCCs (Table 2). Approximately half (48.0%) the
surgeries were performed in Seoul, which showed the highest relevance index (92.6%). The relevance
index for Busan and the surrounding area was 75.2% (Figure 2A). Forty-two percent children residing
in Seoul used home healthcare nursing services. On the other hand, <30% children residing distant</li>
from the capital city used home healthcare nursing services (Figure 2B).

**Table 2.** Demographics of the patients who underwent tracheotomy before 2 years of age (n, %)

| Total               |                              | 813 (n) | 100 (%) |
|---------------------|------------------------------|---------|---------|
| Sor                 | Male                         | 461     | 56.7    |
| Sex                 | Female                       | 352     | 43.3    |
| A go at trachastamy | 0 years old                  | 701     | 86.2    |
| Age at tracheostomy | 1 year old                   | 112     | 13.8    |
|                     | Medical aid + first (lowest) | 123     | 15.6    |
| Level of income     | Second                       | 173     | 21.9    |
| Level of income     | Third                        | 286     | 36.2    |
|                     | Fourth (highest)             | 208     | 26.3    |
| Residence           | Metropolitan city*           | 370     | 41.0    |
| Residence           | Other regions                | 533     | 59.0    |

|                          |                                     | Respiratory                           | 441             | 54.2 |
|--------------------------|-------------------------------------|---------------------------------------|-----------------|------|
|                          |                                     | Neurologic and neuromuscular          | 331             | 40.7 |
|                          |                                     | Premature and neonatal                | 274             | 33.7 |
|                          |                                     | Cardiovascular                        | 187             | 23.0 |
|                          |                                     | Metabolic                             | 135             | 16.6 |
| Categories of CCC        |                                     | Gastrointestinal                      | 132             | 16.2 |
|                          |                                     | Other congenital or genetic defect    | 99              | 12.2 |
|                          |                                     | Renal and urologic                    | 28              | 3.4  |
|                          | 20                                  | Malignancy                            | 12              | 1.5  |
|                          |                                     | Haematologic or immunologic           | 11              | 1.4  |
|                          |                                     | Miscellaneous                         | 1               | 0.1  |
|                          |                                     | 0                                     | 70              | 8.6  |
|                          |                                     | 1                                     | 217             | 26.7 |
| Numbers of CCC           |                                     | 2                                     | 260             | 32.0 |
|                          |                                     | ≥3                                    | 266             | 32.7 |
| Two-year mortality       |                                     | 5                                     | 307             | 37.8 |
| Median (interquartil     | Median (interquartile) day to death |                                       |                 | -    |
| 173 Note: Patients who d | ed during hospit                    | alisation for tracheotomy were exclud | (52-313)<br>ed. |      |
| Abbreviations: CCC,      | complex chronic                     | e condition.                          |                 |      |
| *Metropolitan city: S    | oul, Busan, Inch                    | neon, Daegu, Daejeon, Gwangju, Ulsa   | n               |      |
| 176                      |                                     |                                       |                 |      |

- 37
  38 177 Healthcare utilisation after tracheotomy
- Table 3 shows data for patients with at least one service used in the first 2 years after tracheotomy.
  - 179 The spend on medical services was more in the first year than in the second year; the median total
  - 180 medical costs were 14,542 and 6,468 USD per capita, respectively (1 USD = 1,115.7 Korean won).
  - 181 The total hospital stay, number of emergency room visits, and use of home healthcare nursing also
- 48 182 decreased. While 26.0% patients received home healthcare nursing in the first year, only 17.9% did so
- 50 183 in the second year.51

**Table 3.** Healthcare service utilisation per person among patients who received it more than once

| 56 |                                   |                                  |
|----|-----------------------------------|----------------------------------|
| 57 | First year after tracheotomy (n = | Second year after tracheotomy (n |
| 58 | 566)                              | = 507)                           |
| 59 |                                   |                                  |

| 1   |                         |                               |                   |             |                   |                   |            |                |
|---|-------------------------|-------------------------------|-------------------|-------------|-------------------|-------------------|------------|----------------|
| 2<br>B<br>4   |                         |                               | First<br>quartile | median      | Third<br>quartile | First<br>quartile | median     | Third quartile |
| 5   | Medical cost (U         | JS dollar)                    | 5,854             | 14,542      | 36,820            | 2,673             | 6,468      | 16,241         |
| 6<br>7  | Hospitalisation         | Total duration (days)         | 9                 | 23          | 37.5              | 5                 | 10         | 26             |
| 8<br>9  |                         | Number of<br>hospitalisations | 2                 | 3           | 5                 | 2                 | 3          | 4              |
| 10  |                         | Cost                          | 3,644             | 12,680      | 38,728            | 1,695             | 4,941      | 16,102         |
| 11<br>12  | Outpatient clinic       | Number of visits              | 14                | 23          | 30                | 17                | 25         | 34             |
| 13  |                         | Cost                          | 966               | 1,974       | 3,563             | 786               | 1,639      | 3,364          |
| 14  | Emergency room          | Number of visits              | 2                 | 3           | 5                 | 1                 | 2          | 4              |
| 15  | Home healthcare nursing | Numbers                       |                   | 147 (26.0%) | )                 |                   | 91 (17.9%) |                |
| <del>16</del><br>17   | -                       | ued in US dollars. Those      | e admitted 1      |             |                   |                   | · /        |                |
| $\begin{array}{c} 19\\ 20\\ 21\\ 22\\ 23\\ 24\\ 25\\ 26\\ 27\\ 28\\ 29\\ 30\\ 31\\ 32\\ 33\\ 34\\ 35\\ 36\\ 37\\ 38\\ 39\\ 40\\ 41\\ 42\\ 43\\ 44\\ 45\\ 64\\ 47\\ 48\\ 49\\ 50\\ 51\\ 52\\ 53\\ 54\\ 55\\ 56\\ 57\\ 58\\ 9\\ 9\end{array}$ | 187 "Admission."        |                               |                   |             |                   |                   |            |                |

| 188 |                   |  |            |  |  |  |  |
|-----|-------------------|--|------------|--|--|--|--|
| 100 |                   |  |            |  |  |  |  |
| 189 | Causes of hosp    | Causes of hospitalisation after tracheotomy                                  |            |  |  |  |  |
| 190 | Table 4 summa     | rises the top 30 causes of hospitalisation within 2 years after tracheotomy. | Respirator |  |  |  |  |
| 191 | illnesses, such a | as pneumonia (n=116) and asthma (n=82), and neurological illnesses, inclu    | ding       |  |  |  |  |
| 192 | convulsions and   | d epilepsy, were frequent diagnoses for admission. Fourteen of the 30 diagr  | noses were |  |  |  |  |
| 193 | considered AC     | SCs.   |            |  |  |  |  |
| 194 |                   |  |            |  |  |  |  |
| 195 | Table 4. List of  | f the top 30 diagnoses in patients who were readmitted after tracheotomy     |            |  |  |  |  |
|     | ICD-10 code       | Diagnosis  | n          |  |  |  |  |
|     | J18.9             | Pneumonia*   | 116        |  |  |  |  |
|     | J45.9             | Asthma*  | 82         |  |  |  |  |
|     | J21.9             | Acute bronchiolitis*   | 76         |  |  |  |  |
|     | R56.8             | Other and unspecified convulsions*   | 53         |  |  |  |  |
|     | R50.9             | Fever  | 45         |  |  |  |  |
|     | J69.0             | Pneumonitis due to food and vomit*   | 43         |  |  |  |  |
|     | G40,9             | Epilepsy, unspecified*   | 40         |  |  |  |  |
|     | J38.6             | Stenosis of larynx   | 40         |  |  |  |  |
|     | A09.9             | Gastroenteritis and colitis of unspecified origin*                           | 35         |  |  |  |  |
|     | A41.9             | Sepsis, unspecified  | 35         |  |  |  |  |
|     | J20.9             | Acute bronchitis*  | 35         |  |  |  |  |
|     | R06.0             | Dyspnoea   | 30         |  |  |  |  |
|     | K21.9             | Gastro-oesophageal reflux disease without oesophagitis*                      | 26         |  |  |  |  |
|     | N39.0             | Urinary tract infection*   | 25         |  |  |  |  |
|     | I46.0             | Cardiac arrest with successful resuscitation                                 | 22         |  |  |  |  |
|     | R13               | Dysphagia  | 20         |  |  |  |  |
|     | 146.9             | Cardiac arrest, unspecified  | 17         |  |  |  |  |
|     | R060              | Other forms of dyspnoea  | 17         |  |  |  |  |
|     | D65               | Disseminated intravascular coagulation [defibrination syndrome]              | 17         |  |  |  |  |
|     | J39.8             | Other specified diseases of upper respiratory tract                          | 15         |  |  |  |  |
|     | G41.9             | Status epilepticus, unspecified*   | 13         |  |  |  |  |
|     | D50.9             | Iron deficiency anaemia, unspecified*  | 13         |  |  |  |  |
|     | J30.4             | Allergic rhinitis, unspecified   | 13         |  |  |  |  |
|     | K92.2             | Gastrointestinal haemorrhage, unspecified                                    | 12         |  |  |  |  |
|     | R92.2<br>R62.0    |  |            |  |  |  |  |
|     |                   | Delayed milestone  | 10         |  |  |  |  |
|     | R05               | Cough  | 1(         |  |  |  |  |
|     | E44.0             | Moderate protein-energy malnutrition   | 10         |  |  |  |  |
|     | G40.4             | Other generalised epilepsy and epileptic syndromes*                          | 9          |  |  |  |  |
|     | J38.7             | Other diseases of larynx   | 9          |  |  |  |  |

| 2               |           |                 |                        |                         |                             |                       |
|-----------------|-----------|-----------------|------------------------|-------------------------|-----------------------------|-----------------------|
| 3<br>4          |           | J18.0           | Bronchopneumoni        | a unspecified*          |                             | 9                     |
| 5<br>6          | 196       |                 | <u> </u>               |                         | nedical conditions were or  |                       |
| 7               |           |                 |                        |                         |                             |                       |
| 8<br>9          | 197       | * Diagnosis co  | rresponding to ambu    | alatory care sensitiv   | e conditions (ACSCs)        |                       |
| 9<br>10         | 198       |                 |                        |                         |                             |                       |
| 11<br>12        | 199       | Risk factors fo | or mortality           |                         |                             |                       |
| 13              | 199       | KISK lactors in |                        |                         |                             |                       |
| 14<br>15        | 200       | The median tin  | ne to death within 2   | years after tracheote   | omy for 307 patients was    | 144 days (IQR: 52–    |
| 16              | 201       | 313). In multiv | variable analyses, thr | ee CCCs indicated       | the highest likelihood of n | nortality (OR 2.654;  |
| 17<br>18        |           | ,               |                        |                         | C                           |                       |
| 19              | 202       | 95% CI, 1.462-  | –4.819), and two CC    | CCs was also associa    | ated with increased mortal  | ity (OR 1.971; 95%    |
| 20<br>21        | 203       | CI, 1.084–3.58  | 5). Home healthcare    | e nursing was signif    | icantly associated with a r | educed mortality risk |
| 22              | 204       | (OP 0.612.05)   | 5% CI, 0.433–0.869)    | (Table 5)               |                             |                       |
| 23<br>24        | 204       | (OK, 0.015, 95  | 70 CI, 0.455-0.809)    | (1 able 3).             |                             |                       |
| 25              | 205       |                 |                        |                         |                             |                       |
| 26<br><u>27</u> | 206       | Table 5. Assoc  | ciated factors of mor  | tality of patients that | t underwent tracheotomy     | before 2 years old    |
| 28              |           |                 |                        | Number of death         | OR (95% CI)                 | adjusted OR (95% CI)* |
| 29<br>2101.um   | ber of C  |                 |                        | (n, %)                  | ` ´ ´                       |                       |
| 31              |           |                 | 0                      | 19 (27.1)               | Reference                   | Reference             |
| 32              |           |                 | 1                      | 62 (28.6)               | 1.074 (0.587–1.963)         | 1.151 (0.621–2.133)   |
| 33              |           |                 | 2                      | 102 (39.2)              | 1.732 (0.967–3.102)         | 1.971 (1.084–3.585)   |
| 34<br>35        |           |                 | ≥3                     | 124 (46.6)              | 2.343 (1.313–4.182)         | 2.654 (1.462–4.819)   |
| 3Sex            |           |                 | Female                 | 137 (38.9)              | Reference                   | Reference             |
| 37<br><u>38</u> |           |                 | Male                   | 170 (36.9)              | 0.917 (0.689–1.220)         | 0.985 (0.730–1.331)   |
| 39ge            | at trach  | eotomy          | 0 years old            | 265 (37.8)              | Reference                   | Reference             |
| 40              |           |                 | 1 year old             | 42 (37.5)               | 0.987 (0.654–1.490)         | 0.937 (0.602–1.457)   |
|                 | el of inc | ome             | Medical aid &<br>1st   | 58 (47.2)               | 1.291 (0.824–2.023)         | 1.209 (0.759–1.924)   |
| 43<br>44        |           |                 | 2nd                    | 59 (34.1)               | 0.749 (0.493–1.138)         | 0.688 (0.448–1.058)   |
| 45              |           |                 | 3rd                    | 97 (33.9)               | 0.743 (0.513–1.074)         | 0.684 (0.467–1.002)   |
| 46<br>47        |           |                 | 4th                    | 85 (40.9)               | Reference                   | Reference             |
| 4 <b>B</b> esi  | dence     |                 | Metropolitan           | 123 (37.2)              | Reference                   | Reference             |
| 49              |           |                 | area                   | 125 (57.2)              |                             |                       |
| 50<br>51        |           |                 | Non-<br>metropolitan   | 183 (38.2)              | 1.045 (0.783–1.396)         | 0.983 (0.725–1.333)   |
| 52              |           |                 | area                   | 105 (50.2)              | 1.045 (0.765–1.576)         | 0.765 (0.725-1.555)   |
| 5Hon            | ne health | ncare nursing   | Yes                    | 67 (30.9)               | 0.663 (0.476-0.923)         | 0.613 (0.433–0.869)   |
| 54<br>55        |           |                 | No                     | 240 (40.3)              | Reference                   | Reference             |
| 56              | 207       |                 |                        |                         |                             |                       |

Note: Patients who died during hospitalisation for tracheotomy were excluded. 

58 Abbreviations: OR: odds ratio; CI: confidence interval; CCC: complex chronic condition 

| 1<br>2<br>3           |                   |  |  |  |  |  |  |
|-----------------------|-------------------|--|--|--|--|--|--|
| 5<br>4<br>5<br>6<br>7 | 210<br>211<br>212 | * Adjusted with number of CCC, sex, age at tracheotomy, level of income, residence, and home health care nursing |  |  |  |  |  |
| 8<br>9                | 213               | DISCUSSION   |  |  |  |  |  |
| 10<br>11              | 214               | To our knowledge, this is the first study that identified the nature of healthcare utilisation and               |  |  |  |  |  |
| 12<br>13<br>14<br>15  | 215               | outcomes of children who underwent tracheotomy before 2 years of age in Korea. The number of                     |  |  |  |  |  |
|                       | 216               | infants requiring tracheotomy increased despite a declining national birth rate. Most paediatric                 |  |  |  |  |  |
| 16<br>17              | 217               | patients underwent surgery in their first year of life and had at least one CCC. Presence of more CCCs           |  |  |  |  |  |
| 18<br>19<br>20        | 218               | was associated with higher mortality within 2 years after surgery. Home healthcare nursing utilisation           |  |  |  |  |  |
| 20<br>21<br>22        | 219               | was associated with lower mortality.   |  |  |  |  |  |
| 23<br>24              | 220               | The increasing trend of tracheotomies observed in this study has also been observed in other                     |  |  |  |  |  |
| 25<br>26              | 221               | studies. A single-tertiary centre study revealed that surgeries have been increasing over the last 30            |  |  |  |  |  |
| 27<br>28<br>29<br>30  | 222               | years [4]. A study of 14,155 participants registered in the Pediatric Health Information System                  |  |  |  |  |  |
|                       | 223               | database of 52 children's hospitals in the USA from 2010 to 2018 also showed an increase in the                  |  |  |  |  |  |
| 31<br>32              | 224               | annual number of tracheotomies [16]. This tendency could be attributed to improvements in paediatric             |  |  |  |  |  |
| 33<br>34              | 225               | critical care technologies and the increased life expectancy of medically complex children [1].                  |  |  |  |  |  |
| 35<br>36<br>37        | 226               | Children who underwent tracheotomy had higher healthcare utilisation than did the general                        |  |  |  |  |  |
| 38<br>39              | 227               | population of children. The median total admission durations were 23 and 10 days in the first and                |  |  |  |  |  |
| 40<br>41              | 228               | second years after tracheotomy, respectively; these were substantially higher than reported medians of           |  |  |  |  |  |
| 42<br>43              | 229               | 6.8 and 7.5 days, respectively, for same-age Korean children [17]. The annual median total medical               |  |  |  |  |  |
| 44<br>45              | 230               | costs per capita in both years were also far above those for same-age Korean children (917.5 USD)                |  |  |  |  |  |
| 46<br>47              | 231               | [17]. As the benefit coverage rate in Korea was approximately 60% during the study period, higher                |  |  |  |  |  |
| 48<br>49              | 232               | actual medical expenses were estimated [18]. These results were consistent with previous findings                |  |  |  |  |  |
| 50<br>51              | 233               | [19, 20]. A study including 502 children in the USA who underwent tracheotomy in 2009 found that                 |  |  |  |  |  |
| 52<br>53              | 234               | the total healthcare spending for hospitalisation during 2 years after the surgery was over 75,000 US            |  |  |  |  |  |
| 54<br>55              | 235               | dollars [21].  |  |  |  |  |  |
| 56<br>57<br>58        | 236               | In the current study, approximately 48.0% patients underwent surgery in Seoul, and the                           |  |  |  |  |  |
| 59<br>60              | 237               | relevance index for other regions was lower than that for the capital city. Moreover, the home nursing           |  |  |  |  |  |
|                       |                   |  |  |  |  |  |  |

14

care utilisation rate was higher in Seoul. Patients living in remote areas of the capital city may have difficulties to not only undergo surgery but also receive postoperative supportive care. Studies have urged the establishment of a well-equipped environment and a partnership between hospitals and community care systems for successful tracheostomy management after discharge [22, 23]. To provide proper care after discharge, a referral system and home health care services must be developed throughout the country. For example, the American Academy of Pediatrics has called for fostering family provider-community partnerships [24]. Establishment and implementation of comprehensive measures, such as standardised education protocols, systematic communication between tertiary care centres and primary care providers, and activation of visiting home healthcare, respite care, and school-based specialised services, are encouraged [25]. Most hospitalisations after tracheotomy in this study were due to respiratory or neurological conditions. Especially, among the top 10 conditions, seven were identified as ACSCs. Because admissions due to ACSCs are preventable, increasing parental education and medical accessibility could help minimise avoidable hospitalisations [12, 13]. A survey reported that caregivers of medical technology-dependent children in Korea had substantial problems taking care of their sick children and managing medical devices [26]. The American Thoracic Society guidelines emphasise the importance of education. Continuous efforts to obtain, strengthen, and improve skills are encouraged to ensure patient safety and clinical benefits. The guidelines presented detailed educational objectives in various domains [27]. The utilisation of telemedicine can be an additional option for supporting parents, especially those with limited medical resources. Twenty-seven percent of hospitalisations were shown to be prevented by telemedicine in a prospective clinical study of tracheostomy-dependent children, with the caregivers reporting improved safety and quality of life [28]. In this study, the mortality rate within 2 years after surgery was 37.8%, higher than that previously reported. A retrospective single-centre study including 68 children who underwent tracheotomy before age 2 found that 23.5% patients died [29]. Other recent studies reported that approximately 13%–27% paediatric patients who underwent tracheotomy died [30–33]. The higher mortality rate may be attributed to the broader indications for tracheotomy in Korea, where doctors 

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sometimes perform the procedure even when it may not significantly alter the prognosis. This can be inferred from the higher population-based tracheotomy incidence rate in Korea (10.9 per 100,000 child-years vs. 6.0 per 100,000 child-years in the USA) and deserves further study [6]. To minimise avoidable tracheotomies in infants, a multidisciplinary approach involving evidence-based guideline development, specialised training for healthcare professionals, clear communication with families about prognosis and treatment options, and early discussions on advanced care planning is crucial. Additionally, regular case reviews and data analysis can contribute to improved decision-making and outcomes. Furthermore, those receiving home nursing care demonstrated lower odds of mortality in this study. Establishing supportive systems like home healthcare could also potentially contribute to reduced mortality rates.

This study had limitations. The KNHIS is based on claims data and we could not obtain detailed clinical data or identify the patient's medical condition. Although we used CCC codes to identify the disease severity, it may not have reflected the patient's actual medical condition. Moreover, we could not ascertain the specific reasons for tracheotomy or underlying causes of mortality. Therefore, prospective cohort studies should be conducted to determine pre- and postoperative conditions and prognoses in the future. Nevertheless, this study had several strengths. First, it was a nationwide study that identified children who underwent tracheotomy within 2 years after birth. Because 97% individuals are covered by a unified national health insurance system [34], our findings could be generalised to the entire population and minimise bias. Second, because the KNHIS database contains healthcare utilisation variables, a nationwide analysis of healthcare expenditure and admission days was possible. Thus, our study can be used as a reference for preoperative counselling and parental preparation during the postoperative phase. In conclusion, children with tracheostomies often experience complex conditions. Traditional healthcare models have difficulty meeting the high healthcare needs of these patients, and they frequently receive fragmented and disorganised care [35]. An integrated care system that links

290 hospital-based specialists with community-based healthcare can be helpful. It is necessary to continue

studying the characteristics, needs, and outcomes of this population, as the information gathered will

| 2<br>3   |     |  |  |  |  |
|--|-----|--|--|--|--|
| 4<br>5   | 292 | be beneficial to patients and caregivers.  |  |  |  |
| 6<br>7   | 293 |  |  |  |  |
| 8<br>9<br>10<br>11<br>12<br>13<br>14<br>15<br>16<br>17<br>18<br>19<br>20<br>21<br>22<br>23<br>24<br>25<br>26<br>27<br>28<br>29<br>30<br>31<br>32<br>33<br>34<br>35<br>36<br>37 | 294 | Acknowledgements   |  |  |  |
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|  | 300 | Research ethics approval   |  |  |  |
|  | 301 | No Ethics Approval   |  |  |  |
|  | 302 |  |  |  |  |
|  | 303 | Competing interests  |  |  |  |
|  | 304 | The authors declare no conflict of interest.   |  |  |  |
|  | 305 |  |  |  |  |
|  | 306 | Author's contribution  |  |  |  |
|  | 307 | Conceptualization: Kim MS, Song IG, Kim YS. Data curation: Song IG, Cho Y-M, Lim Y. Formal     |  |  |  |
| 38<br>39   | 308 | analysis: Cho Y-M, Lim Y. Investigation: Kim MS, Song IG, Kim YS, Lee JW. Methodology: Kim     |  |  |  |
| 40<br>41   | 309 | MS, Song IG, Kim YS, Cho Y-M, Lim Y. Software: Song IG, Cho Y-M, Lim Y. Validation: Song IG,   |  |  |  |
| 42<br>43   | 310 | Cho Y-M, Lim Y. Visualization: Song IG, Cho Y-M, Lim Y. Writing - original draft: Song IG, Kim |  |  |  |
| 44<br>45   | 311 | YS. Writing - review & editing: Kim MS, Song IG, Kim YS, Lee JW, Kwon SK, Suh DI, Park JD.     |  |  |  |
| 46<br>47   | 312 |  |  |  |  |
| 48<br>49   | 313 | Data sharing statement   |  |  |  |
| 50<br>51   | 314 | De-identified individual participant data will not be made available.                          |  |  |  |
| 52<br>53<br>54   | 315 |  |  |  |  |
| 55<br>56   |     |  |  |  |  |
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| 59<br>60   |     |  |  |  |  |
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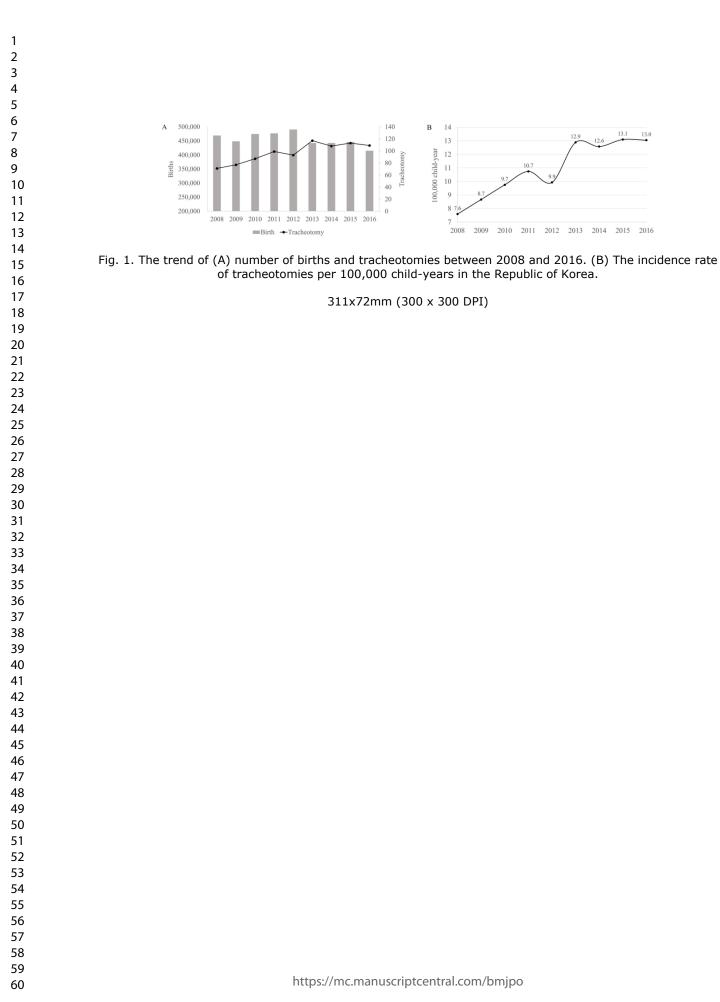
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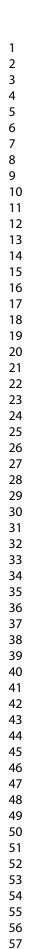
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**Figure Legends** 

Fig. 1. The trend of (A) number of births and tracheotomies between 2008 and 2016. (B) The incidence rate of tracheotomies per 100,000 child-years in the Republic of Korea. Fig. 2. (A) Relevance index (B) home healthcare nursing utilisation ratio for children who had a tracheotomy in the Republic of Korea. Note: The relevance index is the ratio of children who underwent tracheotomy in a certain residential district to children dwelling in that area who underwent tracheotomy. The home healthcare nursing utilisation ratio was defined as the ratio of children utilising home healthcare nursing. The national territory is divided into eight areas. The map was downloaded from the Republic of Korea editable -south-r. .ming the "Terms map template (https://yourfreetemplates.com/free-south-korea-editable-map) and modified by the Microsoft PowerPoint 2016 program after confirming the "Terms-of-use"





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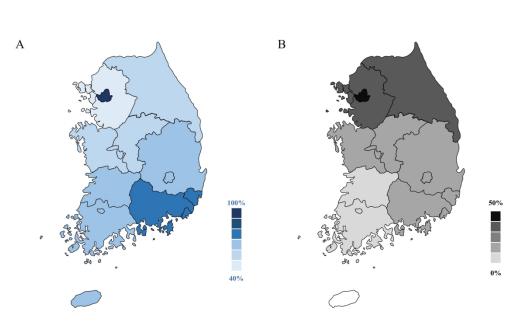


Fig. 2. (A) Relevance index (B) home healthcare nursing utilisation ratio for children who had a tracheotomy in the Republic of Korea.

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# Healthcare service use and medical outcomes of tracheostomy-dependent children: a nationwide study

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Healthcare service use and medical outcomes of tracheostomy-dependent children: a nationwide study

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15 ABSTRACT

Background: Despite the rising trend of tracheostomies in children, there is a lack of
comprehensive resources for families to navigate the challenges of living with a tracheostomy,
emphasising the need for evidence-based support in understanding post-operative care and long-term
adjustments. This study aimed to examine the pattern of utilising healthcare services and nationwide
medical outcomes in children who underwent a tracheotomy before the age of 2 years.

Methods: This retrospective study used the National Health Insurance System database from 2008 to 2016 and included all children codified with tracheotomy procedure codes before their second 24 birthday. Healthcare utilisation, such as medical costs, the number of hospital visits, home healthcare 25 nursing, and the medical diagnoses upon readmission in the first 2 years after tracheotomy, was 26 evaluated. Multivariable logistic regression analysis was used to determine the factors affecting 27 mortality.

Results: In total, 813 patients were included in this study. Their use of healthcare services
and the accompanying expenses were higher than the national medians for similar age groups;
however, both metrics decreased in the second year. The major causes of admission within 2 years of
surgery were respiratory and neurological diseases. The mortality rate within 2 years was 37.8%.
Higher risks of mortality were associated with having two or more complex chronic conditions. Use
of home healthcare nursing services was associated with a lower mortality risk.

34 Conclusion: Paediatric patients with more complex chronic conditions tended to have higher
35 mortality rates within 2 years after surgery. However, receiving home healthcare nursing was
36 significantly associated with a reduced risk of death. Many causes of hospitalisation may be
37 preventable with education and supportive care. Therefore, further research for establishing an
38 integrated care system for these patients and their caregivers is required.

40 Keywords: Tracheotomy; Paediatrics; Health Services; Home Care Services; Mortality
41

42 Key Messages

| 1<br>2   |    |   |
|--|----|---|
| 2<br>3<br>4  | 43 | What is already known on this topic   |
| 5  | 44 | Children under the age of 2 who undergo tracheotomy experience high morbidity, significant            |
| 7<br>8   | 45 | complications, and place a substantial burden on their families.                                      |
| 9<br>10  | 46 |   |
| 11<br>12   | 47 | What this study adds  |
| 13<br>14   | 48 | Many causes of hospitalisation may be preventable with education and supportive care. Receiving       |
| 15<br>16   | 49 | home healthcare nursing was significantly associated with a reduced risk of mortality.                |
| 17<br>18   | 50 |   |
| 19<br>20   | 51 | How this study might affect research, practice or policy  |
| 21<br>22<br>23   | 52 | The findings of this study can serve as empirical evidence for establishing new systems, such as home |
| 24<br>25   | 53 | healthcare services, in countries with weak support systems for children who undergo tracheotomy.     |
| 27<br>28<br>29<br>30<br>31<br>32<br>33<br>34<br>35<br>36<br>37<br>38<br>39<br>40<br>41<br>42<br>43<br>44<br>45<br>46<br>47<br>48<br>49<br>50<br>51<br>52<br>53<br>54<br>55<br>56<br>57<br>58<br>59<br>60 |    | healthcare services, in countries with weak support systems for children who undergo tracheotomy.     |

#### 55 INTRODUCTION

With the development of medical science, the number of medically complex and technology-dependent children has steadily increased. Paediatric tracheostomy is technically difficult, with a higher risk of mortality and perioperative complications than that in the adult population [1, 2]. Particularly for younger children. Data from the National Surgical Quality Improvement Program of the American College of Surgeons indicate that children aged <2 years with tracheostomy have higher morbidity, with 24.3% experiencing a major complication within 30 days of tracheotomy [3]. Nonetheless, the procedure is becoming more prevalent in paediatric populations with complex medical conditions or chronic illnesses, such as airway obstruction, cardiopulmonary disease, and neurological impairment [1, 2, 4].

Living with a tracheostomy poses significant challenges for both patients and their families; therefore, they should receive comprehensive information before the procedure. Providing practical insights into the journey after tracheostomy can empower parents to make informed decisions about the surgery and prepare for the required long-term adjustments. It becomes imperative to develop family-centred resources grounded in robust, evidence-based data on contemporary tracheotomy-related healthcare and outcomes. While there is a wealth of literature on paediatric tracheotomy procedures, indications, and complications, studies focusing on the enduring experiences of patients and their families after tracheotomy are lacking [1, 5-7]. This study leveraged the Korean National Health Insurance System (KNHIS) database to examine nationwide healthcare utilisation and outcomes among children who underwent tracheotomy before their second birthday.

**METHODS** 

#### 77 Data source and study design

This retrospective study analysed the data of children born between 2008 and 2016 using a nationwide
database from the Republic of Korea (Korea) and followed their records until 2018. The KNHIS
database collects mandatory information on beneficiaries of national health insurance and medical aid
in a unified manner. This database comprises several linked datasets such as social and economic

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qualifications, medical treatment, results of medical check-ups, and costs [8]. Diagnostic codes are based on the World Health Organization's International Statistical Classification of Disease and Related Health Problems 10<sup>th</sup> revision (ICD-10). Information on the population census and national annual medical fees was obtained from the Korean Statistical Information Service [9]. The Institutional review board of Seoul National University Hospital reviewed and exempted this study from formal approval as the KNHIS database does not contain any identifiable information (No. 2208-153-1353). The study was conducted according to the principles of the Declaration of Helsinki. Children who underwent tracheotomy before the age of 2 were included to focus on a population where tracheotomy complications are more prevalent [3]. We excluded children who died during hospitalisation to ensure that the study assessed progress after elective tracheotomy, which would be more informative in terms of long-term implications. Terminology Tracheotomy cases were defined as those with a corresponding procedural code (O1300, O1301, O1303, O1305, O1306, M5830). Complex chronic conditions (CCCs) are defined as "any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or one organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center" [10]. The list of categories was adopted from CCC version 2 [11]. Patients living with CCCs were defined as individuals assigned CCC disease codes in either the primary or additional diagnosis field for hospital visits. The number of CCCs was defined as the number of CCC categories designated for each patient. Ambulatory care sensitive conditions (ACSCs) refer to clinical conditions wherein the likelihood of an unplanned hospitalisation can be diminished through prompt and efficient outpatient care. Hospitalisation stemming from ACSC may signify a lost opportunity for prevention and an adverse encounter for a child and their family. Additionally, it may signal a deficiency in, or limited access to, high-quality outpatient healthcare services. We defined ACSCs using the patients' ICD-10

109 codes, according to previous studies [12, 13].

110 The relevance index indicates the number of patients in a certain area who used hospitals in 111 the same region. This was calculated by dividing the number of patients who underwent tracheotomy 112 in a hospital located in the same residential area by the total number of patients who underwent 113 tracheotomy residing in that area for the same year. Therefore, a lower relevance index in a specific 114 region indicated that more patients moved to a remote area for treatment [14, 15].

116 Variables

Demographic characteristics included patients' sex, age at tracheotomy, income level, categories and number of CCCs, address of the patient's residence, and the medical institution where tracheotomy was performed. The insurance type (national health insurance or medical aid) and the NHIS premium based on income levels were utilised as proxy indicators for the financial status. The lowest income category was designated as those receiving medical aid. KNHIS income levels were divided into four groups: category 1 (<25% premium); category 2 (25%-50% premium); category 3 (50%-75% premium); and category 4 (>75% premium). Medical aid and category 1 patients were merged for analysis because of the small number receiving medical aid. The country was divided into administrative districts, and patients' residential districts were classified as metropolitan (Seoul, Busan, Incheon, Daegu, Daejeon, Gwangju, Ulsan) or other regions. For 2 years following tracheotomy, healthcare utilisation indicators included medical expenses and the combined count of hospitalisations, outpatient visits, and use of emergency medical 

services. Notably, admissions through the emergency room were recorded under hospitalisations inthe database, precluding a separate tally for emergency department visits. Additionally, home care

131 nursing utilisation was identified by assignment of procedure codes (AN200, AN300, AN400,

132 AN500).

**Outcome measures** 

135 The main outcome measure was mortality within 2 years of tracheotomy. The primary and top 5

| 2              |     |   |  |  |  |
|----------------|-----|---|--|--|--|
| 3<br>4         | 136 | secondary diagnoses for admission were identified using ICD-10 diagnostic codes. The top 20 codes       |  |  |  |
| 5<br>6         | 137 | were selected after excluding codes relating to health status/services (tracheostomy status) and        |  |  |  |
| 7<br>8         | 138 | underlying medical conditions, such as hypoxic ischemic encephalopathy.                                 |  |  |  |
| 9<br>10        | 139 |   |  |  |  |
| 11<br>12       | 140 | Statistical analysis  |  |  |  |
| 13<br>14       | 141 | The annual difference in the 2-year mortality rate after tracheotomy was examined using Pearson's       |  |  |  |
| 15<br>16       | 142 | chi-square test. Multivariate logistic regression was used to evaluate the association between the main |  |  |  |
| 17<br>18<br>10 | 143 | outcome and predictive factors. All analyses were performed using SAS software (version 9.4; SAS        |  |  |  |
| 19<br>20<br>21 | 144 | Institute Inc., Cary, NC, USA). P values of <0.05 were considered statistically significant, and odds   |  |  |  |
| 22<br>23       | 145 | ratios (ORs) with 95% confidence intervals (CIs) were calculated to elucidate the strengths of the      |  |  |  |
| 24<br>25       | 146 | associations.   |  |  |  |
| 26<br>27       | 147 |   |  |  |  |
| 28<br>29       | 148 | Patient and public involvement  |  |  |  |
| 30<br>31       | 149 | The public nor patients were involved in this study.  |  |  |  |
| 32<br>33       | 150 |   |  |  |  |
| 34<br>35       | 151 | RESULTS   |  |  |  |
| 36<br>37       | 152 | Trends in the number of tracheotomies and mortality rates   |  |  |  |
| 38<br>39       | 153 | In total, 4,105,326 infants were born between 2008 and 2016, with a downward trend (Figure 1A,          |  |  |  |
| 40<br>41<br>42 | 154 | Table 1). However, the number and incidence rate of tracheotomies in patients <2 years also increased   |  |  |  |
| 42<br>43<br>44 | 155 | over the years (Figure 1B). Sixty-one children who died during hospitalisation for tracheotomy were     |  |  |  |
| 45<br>46       | 156 | excluded, and 813 were included. Three hundred and seven patients died within 2 years after             |  |  |  |
| 47<br>48       | 157 | tracheotomy. The overall mortality rate was 37.8%. There were no significant annual differences in      |  |  |  |
| 49<br>50       | 158 | the 2-year mortality rates (P=0.099).   |  |  |  |
| 51<br>52       | 159 |   |  |  |  |
| 53<br>54<br>55 | 160 | Table 1. Number of births and tracheotomies between 2008 and 2016 in the Republic of Korea              |  |  |  |

|  | Birth (n) | Tracheotomy<br>(n) | Death within<br>tracheotomy<br>episode* (n, %) | Death within 2 years<br>after 1 <sup>st</sup> discharge (n, %) |
|--|-----------|--------------------|--|--|
|--|-----------|--------------------|--|--|

| 2008  | 469,248   | 71  | 5 (7.0)   | 24 (36.4)  |
|-------|-----------|-----|-----------|------------|
| 2009  | 448,459   | 77  | 10 (13.0) | 26 (38.8)  |
| 2010  | 474,435   | 87  | 3 (3.4)   | 43 (51.2)  |
| 2011  | 476,710   | 99  | 5 (5.1)   | 29 (30.9)  |
| 2012  | 490,472   | 93  | 3 (3.2)   | 31 (34.4)  |
| 2013  | 442,418   | 117 | 6 (5.1)   | 36 (32.4)  |
| 2014  | 442,341   | 108 | 9 (8.3)   | 36 (36.4)  |
| 2015  | 446,160   | 113 | 8 (7.1)   | 36 (34.3)  |
| 2016  | 415,083   | 109 | 12 (11.0) | 46 (47.4)  |
| Total | 4,105,326 | 874 | 61 (7.0)  | 307 (37.8) |

\* Number of children who died during hospitalisation for tracheotomy and these were excluded in this study.

**Demographic characteristics** 

Most patients underwent tracheotomy before 1 year of age. The majority (91.4%) had at least one
CCC, and approximately two-thirds had multiple CCCs (Table 2). Approximately half (48.0%) the
surgeries were performed in Seoul, which showed the highest relevance index (92.6%). The relevance
index for Busan and the surrounding area was 75.2% (Figure 2A). Forty-two percent children residing
in Seoul used home healthcare nursing services. On the other hand, <30% children residing distant</li>
from the capital city used home healthcare nursing services (Figure 2B).

**Table 2.** Demographics of the patients who underwent tracheotomy before 2 years of age (n, %)

| Total               |                              | 813 (n) | 100 (%) |
|---------------------|------------------------------|---------|---------|
| <i>a</i>            | Male                         | 461     | 56.7    |
| Sex                 | Female                       | 352     | 43.3    |
| A                   | 0 years old                  | 701     | 86.2    |
| Age at tracheostomy | 1 year old                   | 112     | 13.8    |
|                     | Medical aid + first (lowest) | 123     | 15.6    |
| I                   | Second                       | 173     | 21.9    |
| Level of income     | Third                        | 286     | 36.2    |
|                     | Fourth (highest)             | 208     | 26.3    |
| Residence           | Metropolitan city*           | 370     | 41.0    |

| 1<br>2<br>3<br>4<br>5<br>6  |  |
|---|--|
| 7<br>8<br>9<br>10<br>11<br>12<br>13<br>14<br>15<br>16<br>17<br>18<br>19 |  |
| 14<br>15<br>16<br>17<br>18<br>19<br>20                                  |  |
| 20<br>21<br>22<br>23<br>24<br>25<br>26<br>27<br>28<br>29                |  |
| 30<br>31<br>32<br>33  |  |
| 34<br>35<br>36<br>37<br>38<br>39<br>40<br>41                            |  |
| 42<br>43<br>44<br>45<br>46<br>47  |  |
| 48<br>49<br>50<br>51<br>52<br>53<br>54                                  |  |
| 55<br>56<br>57<br>58<br>59<br>60  |  |

|     |   | Other regions                             | 533             | 59.0         |  |  |
|-----|---|---|-----------------|--------------|--|--|
|     |   | Respiratory                               | 441             | 54.2         |  |  |
|     |   | Neurologic and neuromuscular              | 331             | 40.7         |  |  |
|     |   | Premature and neonatal                    | 274             | 33.7         |  |  |
|     |   | Cardiovascular                            | 187             | 23.0         |  |  |
|     |   | Metabolic                                 | 135             | 16.6         |  |  |
|     | Categories of CCC   | Gastrointestinal                          | 132             | 16.2         |  |  |
|     | No.   | Other congenital or genetic defect        | 99              | 12.2         |  |  |
|     |   | Renal and urologic                        | 28              | 3.4          |  |  |
|     |   | Malignancy                                | 12              | 1.5          |  |  |
|     |   | Haematologic or immunologic               | 11              | 1.4          |  |  |
|     |   | Miscellaneous                             | 1               | 0.1          |  |  |
|     |   | 0   | 70              | 8.6          |  |  |
|     | Numbers of CCC  | 1   | 217             | 26.7         |  |  |
|     |   | 2<br>≥3                                   | 260<br>266      | 32.0<br>32.7 |  |  |
|     | Two-year mortality  | 23  | 307             | 37.8         |  |  |
|     |   |   | 144             | 57.0         |  |  |
| 174 | Median (interquartile) day to death   |   | (52-313)        | -            |  |  |
| 174 | Note: Patients who died during hospitalisation for tracheotomy were excluded.                     |   |                 |              |  |  |
| 175 | Abbreviations: CCC, complex chronic   | e condition.                              |                 |              |  |  |
| 176 | *Metropolitan city: Seoul, Busan, Inch  | eon, Daegu, Daejeon, Gwangju, Ulsa        | n               |              |  |  |
| 177 |   |   |                 |              |  |  |
| 178 | Healthcare utilisation after tracheot   | omy                                       |                 |              |  |  |
| 179 | Table 3 shows data for patients with a  | t least one service used in the first 2 y | ears after trac | cheotomy.    |  |  |
| 180 | The spend on medical services was me  | ore in the first year than in the second  | year; the me    | dian total   |  |  |
| 181 | medical costs were 14,542 and 6,468 USD per capita, respectively (1 USD = 1,115.7 Korean won).    |   |                 |              |  |  |
| 182 | The total hospital stay, number of emergency room visits, and use of home healthcare nursing also |   |                 |              |  |  |
| 183 | decreased. While 26.0% patients recei   | ved home healthcare nursing in the fir    | rst year, only  | 17.9% did so |  |  |
| 184 | in the second year.   |   |                 |              |  |  |
| 185 |   |   |                 |              |  |  |
| 186 | Table 3. Healthcare service utilisation   | per person among patients who recei       | ved it more the | han once     |  |  |

| 1<br>2   |                         |                            |                   |                      |                |                   |                         |                |
|--|-------------------------|----------------------------|-------------------|----------------------|----------------|-------------------|-------------------------|----------------|
| В<br>4<br>5  |                         |                            |                   | r after trached 566) | otomy (n =     | Second yea        | The first trace $= 507$ | neotomy (n     |
| 6<br>7   |                         |                            | First<br>quartile | median               | Third quartile | First<br>quartile | median                  | Third quartile |
| 8  | Medical cost (U         | JS dollar)                 | 5,854             | 14,542               | 36,820         | 2,673             | 6,468                   | 16,241         |
| 10   | Hospitalisation         | Total duration (days)      | 9                 | 23                   | 37.5           | 5                 | 10                      | 26             |
| 11<br>12<br>13   |                         | Number of hospitalisations | 2                 | 3                    | 5              | 2                 | 3                       | 4              |
| 13   |                         | Cost                       | 3,644             | 12,680               | 38,728         | 1,695             | 4,941                   | 16,102         |
| 14<br>15   | Outpatient clinic       | Number of visits           | 14                | 23                   | 30             | 17                | 25                      | 34             |
| 15<br>16   |                         | Cost                       | 966               | 1,974                | 3,563          | 786               | 1,639                   | 3,364          |
| 17   | Emergency room          | Number of visits           | 2                 | 3                    | 5              | 1                 | 2                       | 4              |
|  | Home healthcare nursing | Numbers                    |                   | 147 (26.0%)          | )              |                   | 91 (17.9%)              |                |
| <del>19</del><br>20  | ¥                       | ued in US dollars. Those   | e admitted t      | .1 1                 | ·              |                   | , ,                     |                |
| $\begin{array}{c} 21\\ 22\\ 23\\ 24\\ 25\\ 26\\ 27\\ 28\\ 29\\ 30\\ 1\\ 32\\ 33\\ 35\\ 36\\ 37\\ 38\\ 90\\ 41\\ 42\\ 43\\ 44\\ 56\\ 47\\ 48\\ 9\\ 50\\ 1\\ 52\\ 53\\ 56\\ 57\\ 58\\ 90\end{array}$ | 188 "Admission."        | ued in US dollars. Those   |                   |                      |                |                   |                         |                |

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|----------------|-----|-------------------|---|-------------|
| 3<br>4         | 189 |                   |   |             |
| 5<br>6<br>7    | 190 | Causes of hosp    | bitalisation after tracheotomy  |             |
| 8<br>9         | 191 | Table 4 summa     | rises the top 20 causes of hospitalisation within 2 years after tracheotomy.      | Respiratory |
| 10<br>11       | 192 | illnesses, such a | as pneumonia (n=116) and asthma (n=82), and neurological illnesses, inclu         | uding       |
| 12<br>13       | 193 | convulsions and   | d epilepsy, were frequent diagnoses for admission. Ten of the 20 diagnose         | s were      |
| 14<br>15<br>16 | 194 | considered AC     | SCs.  |             |
| 17<br>18       | 195 |                   |   |             |
| 19<br>20       | 196 | Table 4. List of  | f the top 30 diagnoses in patients who were readmitted after tracheotomy          |             |
| 21             |     | ICD-10 code       | Diagnosis   | n           |
| 22<br>23       |     | J18.9             | Pneumonia*  | 116         |
| 24             |     | J45.9             | Asthma*   | 82          |
| 25             |     | J21.9             | Acute bronchiolitis*  | 76          |
| 26<br>27       |     | R56.8             | Other and unspecified convulsions*  | 53          |
| 27<br>28       |     | R50.9             | Fever   | 45          |
| 29             |     | J69.0             | Pneumonitis due to food and vomit*  | 43          |
| 30             |     | G40,9             | Epilepsy, unspecified*  | 40          |
| 31<br>32       |     | J38.6             | Stenosis of larynx  | 40          |
| 33             |     | A09.9             | Gastroenteritis and colitis of unspecified origin*                                | 35          |
| 34             |     | A41.9             | Sepsis, unspecified   | 35          |
| 35<br>36       |     | J20.9             | Acute bronchitis*   | 35          |
| 30<br>37       |     | R06.0             | Dyspnoea  | 30          |
| 38             |     | K21.9             | Gastro-oesophageal reflux disease without oesophagitis*                           | 26          |
| 39             |     | N39.0             | Urinary tract infection*  | 25          |
| 40<br>41       |     | I46.0             | Cardiac arrest with successful resuscitation                                      | 22          |
| 42             |     | R13               | Dysphagia   | 20          |
| 43             |     | I46.9             | Cardiac arrest, unspecified   | 17          |
| 44<br>45       |     | R060              | Other forms of dyspnoea   | 17          |
| 46             |     | D65               | Disseminated intravascular coagulation [defibrination syndrome]                   | 17          |
| 47             |     | J39.8             | Other specified diseases of upper respiratory tract                               | 15          |
| 48<br>49       | 197 |                   | health status/services and underlying medical conditions were omitted.            |             |
| 49<br>50       |     |                   |   |             |
| 51             | 198 | * Diagnosis con   | rresponding to ambulatory care sensitive conditions (ACSCs)                       |             |
| 52             | 100 |                   |   |             |
| 53<br>54       | 199 |                   |   |             |
| 55<br>56       | 200 | Risk factors fo   | or mortality  |             |
| 57<br>58       | 201 | The median tim    | ne to death within 2 years after tracheotomy for 307 patients was 144 days        | (IQR: 52-   |
| 59<br>60       | 202 | 313). In multive  | ariable analyses, three CCCs indicated the highest likelihood of mortality and 12 | (OR 2.654;  |
|                |     |                   | 12  |             |

- 95% CI, 1.462-4.819), and two CCCs was also associated with increased mortality (OR 1.971; 95%
  - CI, 1.084–3.585). Home healthcare nursing was significantly associated with a reduced mortality risk
- (OR, 0.613; 95% CI, 0.433-0.869) (Table 5).

Table 5. Associated factors of mortality of patients that underwent tracheotomy before 2 years old 

| 13 207 Table 5. 11550                 |                              | <u> </u>                  | at under went tracheotomy | <u> </u>              |
|---------------------------------------|------------------------------|---------------------------|---------------------------|-----------------------|
| 14<br>15                              | Q_                           | Number of death $(n, \%)$ | OR (95% CI)               | adjusted OR (95% CI)* |
| 16 umber of CCC                       | 0                            | 19 (27.1)                 | Reference                 | Reference             |
| 17<br>18                              | 1                            | 62 (28.6)                 | 1.074 (0.587–1.963)       | 1.151 (0.621–2.133)   |
| 19                                    | 2                            | 102 (39.2)                | 1.732 (0.967–3.102)       | 1.971 (1.084–3.585)   |
| 20<br>21                              | ≥3                           | 124 (46.6)                | 2.343 (1.313–4.182)       | 2.654 (1.462–4.819)   |
| 2Sex                                  | Female                       | 137 (38.9)                | Reference                 | Reference             |
| 23<br>24                              | Male                         | 170 (36.9)                | 0.917 (0.689–1.220)       | 0.985 (0.730–1.331)   |
| $2\frac{4}{2}$ ge at tracheotomy      | 0 years old                  | 265 (37.8)                | Reference                 | Reference             |
| 26                                    | 1 year old                   | 42 (37.5)                 | 0.987 (0.654–1.490)       | 0.937 (0.602–1.457)   |
| 27<br>28<br>29<br>29                  | Medical aid &<br>1st         | 58 (47.2)                 | 1.291 (0.824–2.023)       | 1.209 (0.759–1.924)   |
| 30                                    | 2nd                          | 59 (34.1)                 | 0.749 (0.493–1.138)       | 0.688 (0.448–1.058)   |
| 31                                    | 3rd                          | 97 (33.9)                 | 0.743 (0.513–1.074)       | 0.684 (0.467–1.002)   |
| 32<br>33                              | 4th                          | 85 (40.9)                 | Reference                 | Reference             |
| 314 esidence<br>35                    | Metropolitan<br>area         | 123 (37.2)                | Reference                 | Reference             |
| 36<br>37<br>38                        | Non-<br>metropolitan<br>area | 183 (38.2)                | 1.045 (0.783–1.396)       | 0.983 (0.725–1.333)   |
| <sup>39</sup> Home healthcare nursing | Yes                          | 67 (30.9)                 | 0.663 (0.476–0.923)       | 0.613 (0.433–0.869)   |
| 40<br>41                              | No                           | 240 (40.3)                | Reference                 | Reference             |
| 42 208                                |                              |                           | 6                         |                       |

Note: Patients who died during hospitalisation for tracheotomy were excluded. Abbreviations: OR: odds ratio; CI: confidence interval; CCC: complex chronic condition 

\* Adjusted with number of CCC, sex, age at tracheotomy, level of income, residence, and home health care nursing

- DISCUSSION
- To our knowledge, this is the first study that identified the nature of healthcare utilisation and
- outcomes of children who underwent tracheotomy before 2 years of age in Korea. The number of
- infants requiring tracheotomy increased despite a declining national birth rate. Most paediatric
- patients underwent surgery in their first year of life and had at least one CCC. Presence of more CCCs

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was associated with higher mortality within 2 years after surgery. Home healthcare nursing utilisation
was associated with lower mortality.

The increasing trend of tracheotomies observed in this study has also been observed in other studies. A single-tertiary centre study revealed that surgeries have been increasing over the last 30 years [4]. A study of 14,155 participants registered in the Pediatric Health Information System database of 52 children's hospitals in the USA from 2010 to 2018 also showed an increase in the annual number of tracheotomies [16]. This tendency could be attributed to improvements in paediatric critical care technologies and the increased life expectancy of medically complex children [1].

Children who underwent tracheotomy had higher healthcare utilisation than did the general population of children. The median total admission durations were 23 and 10 days in the first and second years after tracheotomy, respectively; these were substantially higher than reported medians of 6.8 and 7.5 days, respectively, for same-age Korean children [17]. The annual median total medical costs per capita in both years were also far above those for same-age Korean children (917.5 USD) [17]. As the benefit coverage rate in Korea was approximately 60% during the study period, higher actual medical expenses were estimated [18]. These results were consistent with previous findings [19, 20]. A study including 502 children in the USA who underwent tracheotomy in 2009 found that the total healthcare spending for hospitalisation during the 2 years after the surgery was over 75,000 US dollars [21].

In the current study, approximately 48.0% patients underwent surgery in Seoul, and the relevance index for other regions was lower than that for the capital city. Moreover, the home care nursing utilisation rate was higher in Seoul. Patients living in remote areas of the capital city may have difficulties with not only undergoing surgery but also receiving postoperative supportive care. A survey reported that caregivers of medical technology-dependent children in Korea had substantial problems taking care of their sick children and managing medical devices at home [22]. Studies have urged the establishment of a well-equipped environment and a partnership between hospitals and community care systems for successful tracheostomy management after discharge [23, 24]. To provide proper care after discharge, a referral system and home health care services must be 

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developed throughout the country. For example, the American Academy of Pediatrics has called for
fostering family provider-community partnerships [25]. Establishment and implementation of
comprehensive measures, such as standardised education protocols, systematic communication
between tertiary care centres and primary care providers, and activation of visiting home healthcare,
respite care, and school-based specialised services, are encouraged [26, 27].

In this study, most hospitalisations after tracheotomy were due to respiratory or neurological conditions. Notably, among the top 10 conditions, seven were identified as ACSCs and eight among the top 20 conditions were respiratory problems. The aforementioned comprehensive measures could help minimise avoidable hospitalisations [12, 13, 26, 27]. In order to reduce respiratory problems at home, caregivers should be educated on adequate secretion management, clean techniques for aspiration, and the importance of vaccination [28]. The American Thoracic Society guidelines also emphasise the importance of education. Continuous efforts to obtain, strengthen, and improve skills are encouraged to ensure patient safety and clinical benefits. The guidelines presented detailed educational objectives in various domains [29]. The utilisation of telemedicine can be an additional option for supporting parents, especially those with limited medical resources. Twenty-seven percent of hospitalisations were shown to be prevented by telemedicine in a prospective clinical study of tracheostomy-dependent children, with caregivers reporting improved safety and quality of life [30]. In this study, the mortality rate within 2 years after surgery was 37.8%, higher than that 

previously reported. A retrospective single-centre study including 68 children who underwent tracheotomy before age 2 found that 23.5% patients died [31]. Other recent studies reported that approximately 13%–27% paediatric patients who underwent tracheotomy died [32–35]. The higher mortality rate may be attributed to the broader indications for tracheotomy in Korea, where doctors sometimes perform the procedure even when it may not significantly alter the prognosis. This can be inferred from the higher population-based tracheotomy incidence rate in Korea (10.9 per 100,000 child-years vs. 6.0 per 100,000 child-years in the USA) and deserves further study [6]. To minimise avoidable tracheotomies in infants, a multidisciplinary approach involving evidence-based guideline development, specialised training for healthcare professionals, clear communication with families 

about prognosis and treatment options, and early discussions on advanced care planning is crucial.
Additionally, regular case reviews and data analysis can contribute to improved decision-making and
outcomes.

The greater the number of CCCs present among patients who underwent tracheotomy, the higher the association with mortality. Since the specific cause of death for each patient was not ascertainable in this study, caution is warranted in the interpretation of our findings. However, our findings could be utilized in shared decision-making when deciding to proceed with tracheotomy. It would be beneficial to share with parents that the risks/burdens of the procedure, particularly the risk of mortality due to underlying conditions, must be considered.

Those receiving home care nursing demonstrated lower odds of mortality in this study. Home care nursing has been shown in previous studies to assist with transitioning safely from the hospital to home and reducing the risk of readmission. Additionally, research has demonstrated its ability to decrease family burnout and enhance the quality of life for children [36-38]. Furthermore, cost-effectiveness analyses have revealed significant cost-saving effects associated with home care nursing [36, 39]. Establishing supportive systems like home healthcare could potentially contribute to reducing mortality rates and alleviating the burden on families.

This study had limitations. Firstly, the KNHIS is based on claims data and we could not obtain detailed clinical data or identify the patient's medical condition. Although we used CCC codes to identify the disease severity, it may not have reflected the patient's actual medical condition. Moreover, we could not ascertain the specific reasons for tracheotomy or underlying causes of mortality. Therefore, prospective cohort studies should be conducted to determine pre- and postoperative conditions and prognoses in the future. Secondly, as a retrospective cohort study, this study examined associations rather than causal relationships, thus precluding the determination of cause-effect relationship. This aspect could also be further addressed in a prospective cohort study. Nevertheless, this study had several strengths. First, it was a nationwide study that identified children who underwent tracheotomy within 2 years after birth. Because 97% of individuals are covered by a unified national health insurance system [40], our findings could be generalised to the entire

| 1<br>2         |     |  |
|----------------|-----|--|
| 3<br>4<br>5    | 300 | population and minimise bias. Second, because the KNHIS database contains healthcare utilisation   |
| 6<br>7         | 301 | variables, a nationwide analysis of healthcare expenditure and admission days was possible. Thus, our  |
| 8<br>9         | 302 | study can be used as a reference for preoperative counselling and parental preparation during the  |
| 10<br>11       | 303 | postoperative phase.   |
| 12<br>13       | 304 | In conclusion, children with tracheostomies often experience complex conditions.   |
| 14<br>15       | 305 | Traditional healthcare models have difficulty meeting the high healthcare needs of these patients, and   |
| 16<br>17       | 306 | they frequently receive fragmented and disorganised care [41]. An integrated care system that links  |
| 18<br>19<br>20 | 307 | hospital-based specialists with community-based healthcare can be helpful. It is necessary to continue   |
| 20<br>21<br>22 | 308 | studying the characteristics, needs, and outcomes of this population, as the information gathered will   |
| 23<br>24       | 309 | be beneficial to patients and caregivers.  |
| 25<br>26       | 310 |  |
| 27<br>28       | 311 | Acknowledgements   |
| 29<br>30       | 312 | None   |
| 31<br>32       | 313 |  |
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| 38<br>39       | 316 | for-profit sectors.  |
| 40<br>41       | 317 | This research received no specific grant from any funding agency in the public, commercial or not-<br>for-profit sectors.<br><b>Research ethics approval</b><br>No Ethics Approval<br><b>Competing interests</b><br>The authors declare no conflict of interest. |
| 42<br>43       | 318 | No Ethics Approval   |
| 44<br>45       | 319 |  |
| 46<br>47       | 320 | Competing interests  |
| 48<br>49       | 321 | The authors declare no conflict of interest.   |
| 50<br>51       | 322 |  |
| 52<br>53<br>54 | 323 | Author's contribution  |
| 55<br>56       | 324 | Conceptualization: Kim MS, Song IG, Kim YS. Data curation: Song IG, Cho Y-M, Lim Y. Formal   |
| 57<br>58<br>59 | 325 | analysis: Cho Y-M, Lim Y. Investigation: Kim MS, Song IG, Kim YS, Lee JW. Methodology: Kim   |
| 60             |     | 17   |

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| 3<br>4<br>5   | 326 | MS, Song IG, Kim YS, Cho Y-M, Lim Y. Software: Song IG, Cho Y-M, Lim Y. Validation: Song IG,   |
| 6<br>7  | 327 | Cho Y-M, Lim Y. Visualization: Song IG, Cho Y-M, Lim Y. Writing - original draft: Song IG, Kim |
| 8<br>9  | 328 | YS. Writing - review & editing: Kim MS, Song IG, Kim YS, Lee JW, Kwon SK, Suh DI, Park JD.     |
| 10<br>11  | 329 |  |
| 12<br>13  | 330 | Data sharing statement   |
| 14<br>15  | 331 | De-identified individual participant data will not be made available.                          |
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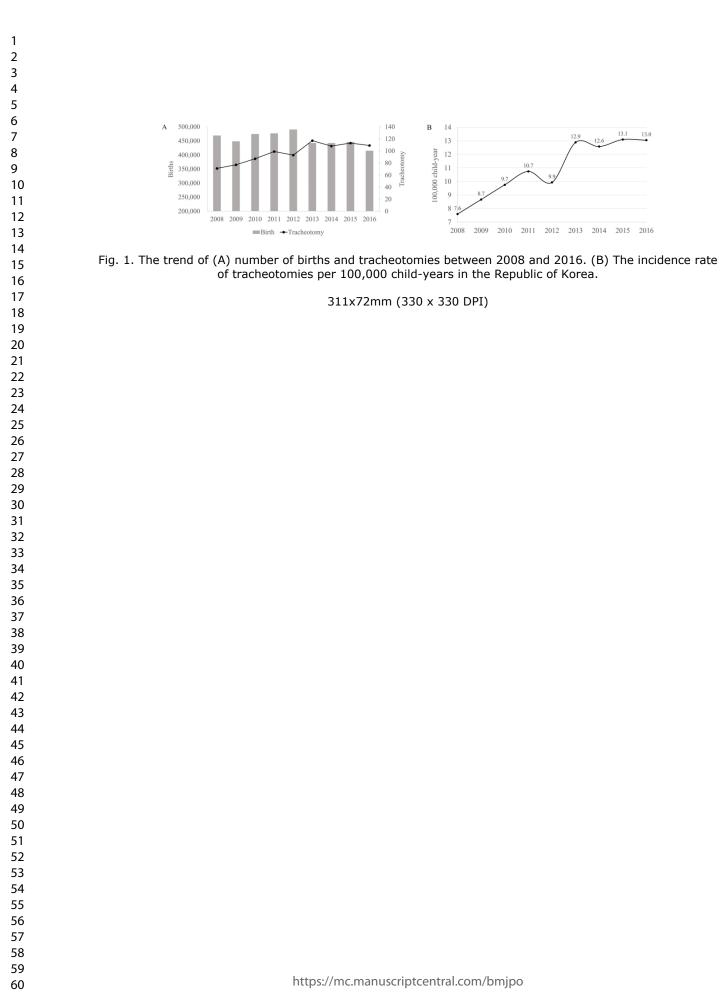
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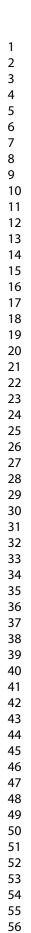
**Figure Legends** 

# 428 Fig. 1. The trend of (A) number of births and tracheotomies between 2008 and 2016. (B) The incidence rate of tracheotomies per 100,000 child-years in the Republic of Korea. 429 430 Fig. 2. (A) Relevance index (B) home healthcare nursing utilisation ratio for children who had a 431 432 tracheotomy in the Republic of Korea. 433 Note: The relevance index is the ratio of children who underwent tracheotomy in a certain residential 434 435 district to children dwelling in that area who underwent tracheotomy. The home healthcare nursing 436 utilisation ratio was defined as the ratio of children utilising home healthcare nursing. The national 437 territory is divided into eight areas. The map was downloaded from the Republic of Korea editable

map template (https://yourfreetemplates.com/free-south-korea-editable-map) and modified by the

439 Microsoft PowerPoint 2016 program after confirming the "Terms-of-use"





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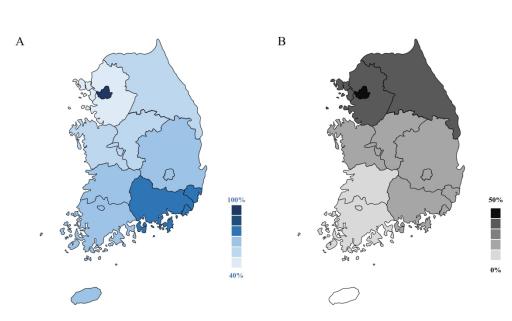


Fig. 2. (A) Relevance index (B) home healthcare nursing utilisation ratio for children who had a tracheotomy in the Republic of Korea.

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