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ORIGINAL ARTICLE

Indications for tracheostomy in children with head and neck lymphatic malformation: analysis of a nationwide survey in Japan

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Abstract

Purpose Airway obstruction caused by lymphatic malformation (LM) in the head and neck may require a tracheostomy. We present the results of our analysis of a nationwide survey on the indications for tracheostomy in children with head and neck LM.

Methods We analyzed data in relation to tracheostomy based on a questionnaire about 518 children with head and neck LM without mediastinal involvement.

Results Tracheostomy was performed for 43 of the 518 children. Most (32/43) of these children were younger than 1 year of age and the tracheostomy was almost always performed for airway obstruction (40/43). The lesion was in contact with the airway in 32 (72%) of these children, but in only 58 (12%) of the 473 children who were managed without tracheostomy. When the maximum circumferential area of contact was compared, only 20 (27%) of 74 patients with maximum contact of less than a half-circle required tracheostomy, whereas 11 of 13 with maximum contact of more than a half-circle required tracheostomy ($P = 0.0001$). Six patients without airway contact required tracheostomy because of acute swelling caused by hemorrhage, infection, or both.

Conclusions Children with head and neck LM required tracheostomy to relieve airway obstruction. Tracheostomy should be considered if the lesion is in contact with the airway and surrounds more than a half-circle, and when it causes acute swelling.

Keywords Lymphangioma · Lymphatic malformation · Neck · Tracheostomy · Management

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Introduction

Lymphangioma or lymphatic malformation (LM) is a rare congenital benign disease caused by the hamartomatous development of the lymphatic vessels. It has been classified by The International Society for the Study of Vascular Anomalies (ISSVA) into common (cystic) LM, generalized lymphatic anomaly (GLA), LM in Gorham–Stout disease (GSD) and "others". LM is further sub-classified into macrocystic, microcystic, and mixed types [1, 2]. The disease appears most commonly in the neck and axilla, sometimes extending into the mediastinum and potentially causing life-threatening symptom(s) from airway obstruction [3–5]. In the Research Project for Intractable Diseases, conducted by the Ministry of Health, Labor and Welfare in Japan, we attempted to draft clinical guidelines for LMs (common LM, GLA, GSD) affecting the airway and other anatomical sites in children. To propose appropriate management for the LM lesion, which is infrequent but presents in various clinical conditions, a nationwide survey to register pediatric cases of LM was carried out by our project team in 2015.

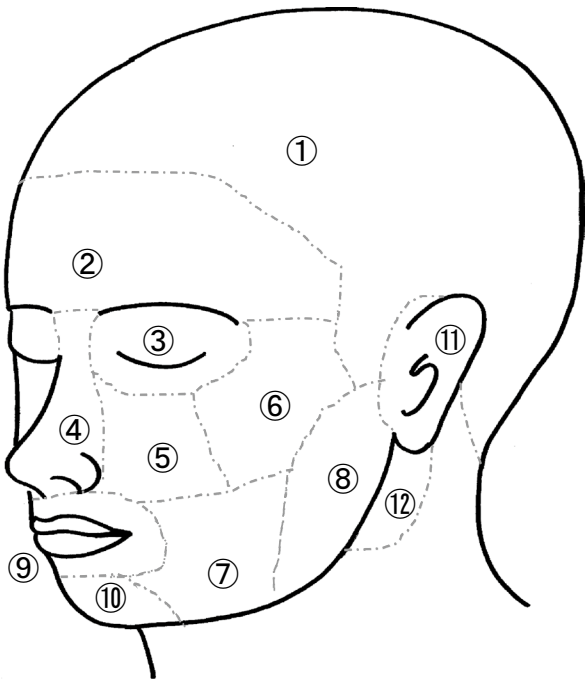
Airway obstruction caused by head and neck LM sometimes requires tracheostomy to maintain the airway integrity and relieve respiratory distress [4, 6]. To secure an airway in a newborn baby, even ex utero intrapartum treatment (EXIT) may be considered when a large head and neck LM is detected prenatally [7–9]. However, childhood tracheostomy has been shown to impair speech development, even after decannulation [10, 11]. Our project composed clinical questions regarding the consideration of tracheostomy for these children. We analyzed the clinical data accumulated by the survey to elucidate the factors that optimize its consideration for a child with a head and neck LM. Children with mediastinal involvement that could affect the airway were also accumulated and their management, including tracheostomy, was discussed in a twin article published recently [12].

Methods

Questionnaire

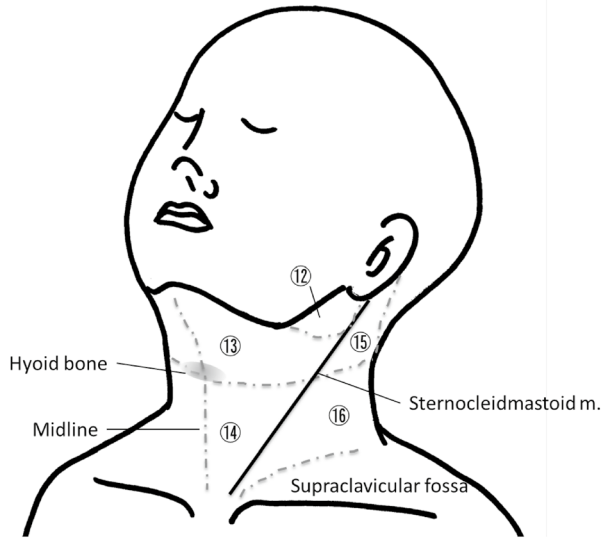
The Research Project for Intractable Diseases conducted by the Ministry of Health, Labor and Welfare tried to establish practical guidelines for treating LM affecting the airway. A nationwide survey to register pediatric patients with LM was carried out by our project team in 2015. The survey was an extensive web-based questionnaire with 273 items on clinical features, including gender, age at onset and diagnosis, perinatal history, symptoms, lesion size and site, radiographic and pathological findings, treatment, complications, clinical course, and outcome. Regarding the lesion site, when LM was located at a site affecting the airway, we asked about the exact anatomical site using figures to define the area (Figs. 1, 2, 3). We also asked about the range and circumference of contact with the airway at four different levels, from the upper pharynx to the intrathoracic trachea, based on radiological images (Fig. 4). Regarding treatment, we asked for extensive details about the tracheostomy indications, including why a tracheostomy was required, the presence or absence of airway obstruction when the tracheostomy was performed, and whether it was temporary with the age of decannulation.

The questionnaire was sent by e-mail to every institute with employed members of the Japanese Society of Pediatric Surgeons. The email requested that respondents access the webpage through the link: http://www.lymphangioma.net/index.html, which was designed to register both patient data and data from the treating surgeon. To avoid reporting



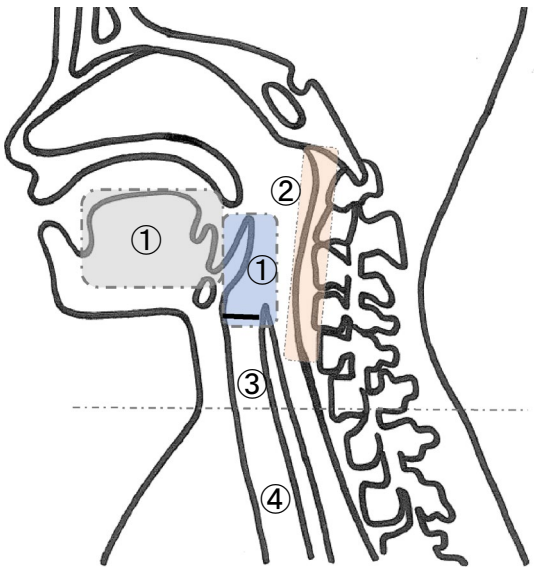
① scalp area
② frontal area
③ orbit area
④ nasal area
⑤ suborbital area
⑥ zygomatic area
⑦ cheek area
⑧ parotid area
⑨ lip area
⑩ mental area
⑪ auricular area
⑫ retromandibular area

Fig. 1 Definition of anatomical sites in the head



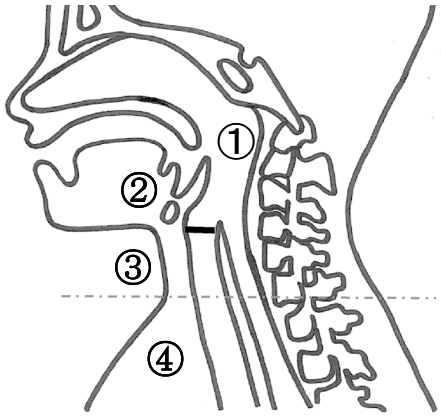
⑫ retromandibular area
⑬ submandibular and submental area (upper anterior triangle)
⑭ anterior neck area (lower anterior triangle)
⑮ lateral neck area (upper posterior triangle)
⑯ lateral neck are (lower posterior triangle)
⑰ posterior neck area (nuchal area)

Fig. 2 Definition of anatomical sites in the neck



① Tongue/Epiglottis/Vocal Cord
② Posterior Pharynx
③ Cervical Trachea
④ Intrathoracic Trachea

Fig. 3 Definition of anatomical sites in the airway



① Upper and middle pharynx (above the epiglottis)
② Larynx (above and below the vocal cord)
③ Cervical trachea
④ Intrathoracic trachea

Fig. 4 Definition of anatomical sites of airway contact

duplicate data, we identified overlapping patients by date of birth, sex, and an endemic number allocated to each patient.

Data analysis and ethical considerations

Statistical analysis was performed with Excel software ystat2002 (Saitama, Japan). Descriptive statistical methods (median and standard deviation) and the Yates chi-square test for two and multi-group comparisons were used for statistical analyses. A value of $P < 0.05$ was considered significant.

The survey involving human participants was conducted in compliance with the ethical standards of the institutional and/or national research guidelines, following the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. The Institutional Review Board of Keio University School of Medicine (20,120,437) and the ethics committee of the Japanese Society of Pediatric Surgeons approved this survey on October 10, 2015. Formal consent is not required for this type of study.

Results

Patients and general characteristics (Figs. 5, 6)

The web-based questionnaire accumulated data on 1718 children with LM, and 606 children with lesions capable of impacting the airway were registered. Among these, 518, with head and neck LM without mediastinal involvement were extracted and the clinical features of each case were

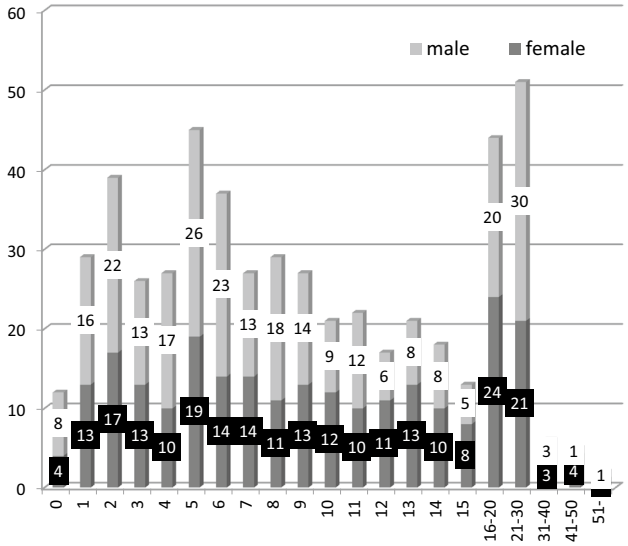


Fig. 5 Gender and age of children at the time of the survey

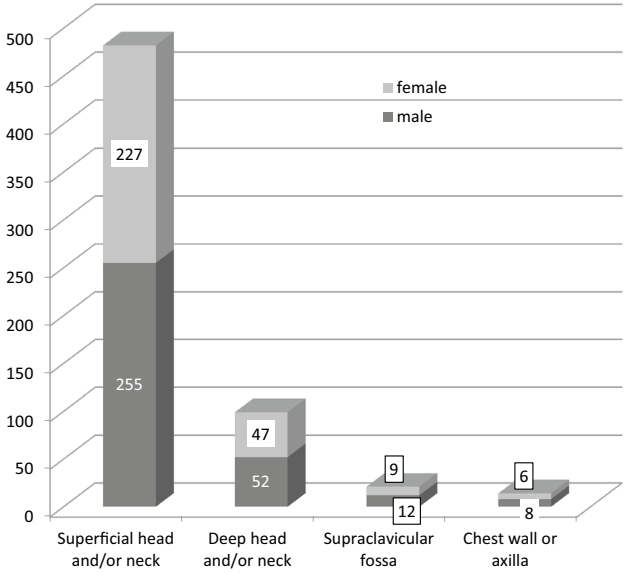


Fig. 7 Sites of the lesion and gender

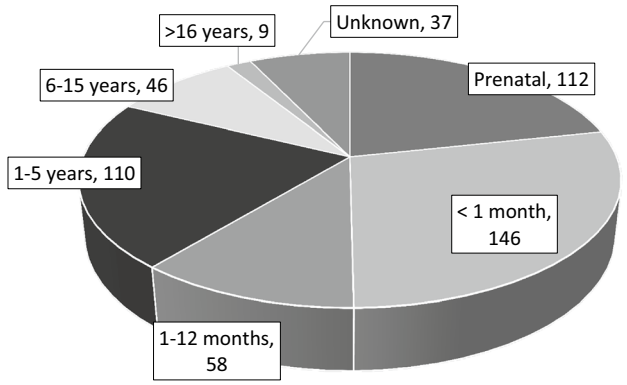


Fig. 6 Age at diagnosis

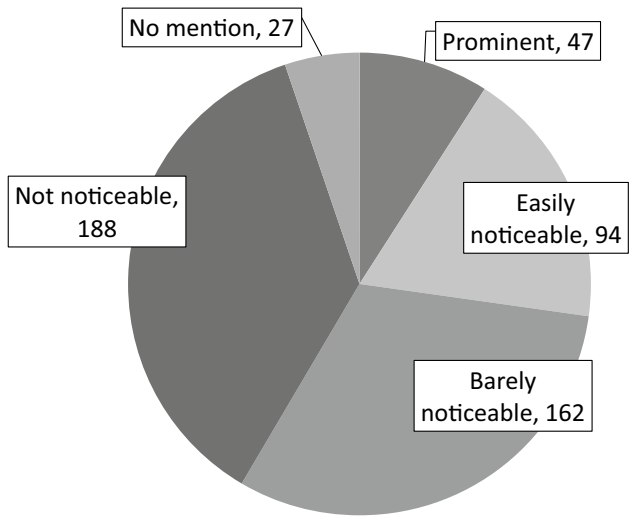


Fig. 8 Disfigurement by the lesion

analyzed for 273 items. Eighty-seven patients with mediastinal involvement were registered, but their clinical features and management, including tracheostomy, were discussed and reported in the twin paper published previously [12]. The male-to-female ratio of children with head and neck LM was 273:245 and the number of patients in each age group, at the time of survey and at the time of diagnosis is shown in Figs. 5 and 6. The lesion was found prenatally in 112 patients (22%) and within the neonatal period in 146 (28%), accounting for 50% of the registered cases, without a significant gender difference (Figs. 5, 6).

Characteristics of head and neck LM (Figs. 7, 8, 9)

The head and neck LMs were located superficially in 482 of the 518 (93%) patients, but some were present in deep sites. The lesion was located either in the oral cavity, pharynx, or larynx in 99 patients (Fig. 7). Many lesions were apparent, but 97 and 47, respectively, were registered as "easily noticeable" or "prominent", accounting for only 27%. The response was "not apparent" or "barely noticeable" for 188 and 162 patients, respectively (Fig. 8) Most lesions were diagnosed as macrocystic LM (cystic lymphangioma) (374/518; 72%), followed by mixed subtype LM (96/518; 19%), and microcystic LM (cavernous lymphangioma) (28/518; 5%). There were only three cases of GLA (Fig. 9).

Airway obstruction (Table 1)

Symptoms of airway obstruction were identified during the clinical course in 73 (14%) of the 518 patients. The survey

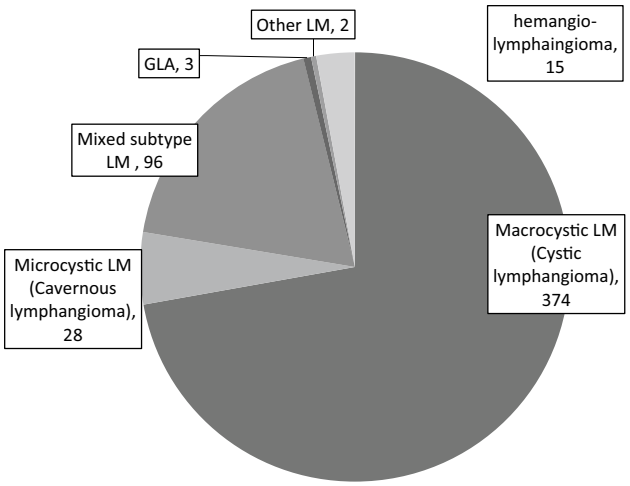


Fig. 9 Classification of the lesion

Table 1 Airway obstruction symptoms during the clinical course

Airway obstruction (+)	Complete obstruction at survey	4 (4*)
	Always stenotic at survey	11 (10*)
	Frequently stenotic at survey	6 (2*)
	Possible at survey	13 (7*)
	No obstruction at survey	39 (17*)
	73 (40*)	
Airway obstruction (−)	443 (3*)	
Unknown	2	
Total	518 (43*)	

*No. of patients who had tracheotomy done during the clinical course

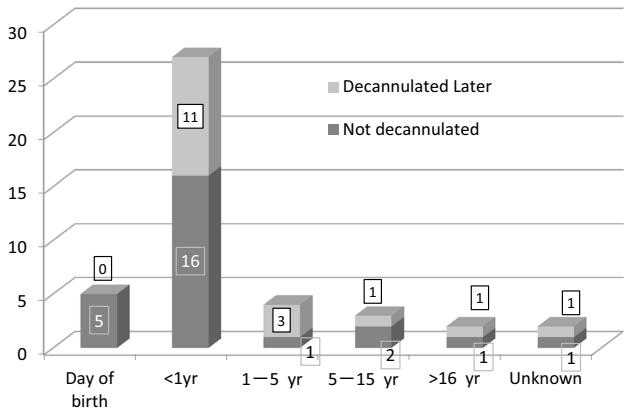


Fig. 10 Age at tracheostomy and decannulation

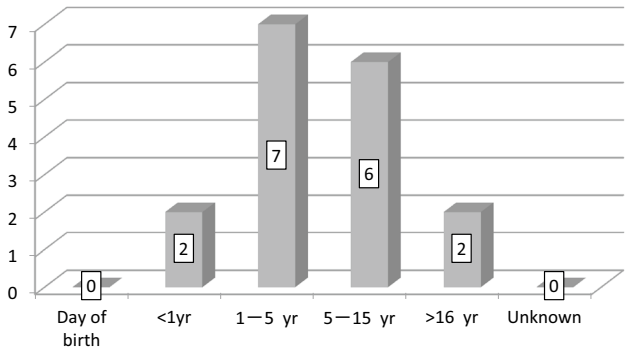


Fig. 11 Age at decannulation

reported vague obstruction symptoms in 34 (6.5%) of the 73 patients, including the answer "possible" in 13. The responses included "completely obstructed" for 4 patients, "always stenotic" for 11, and "frequently stenotic" for 6 (Table 1). Thus, 39 patients who had suffered symptoms were reported as having no symptoms at the time of the survey.

Tracheostomy

Age at tracheostomy and decannulation (Figs. 10, 11)

A tracheostomy was performed for 43 (8.3%) of the 518 patients when most (32/43; 74%) were younger than 1 year of age and it was temporary in 17 (40%) (Fig. 10). Decannulation usually took place between 1 and 15 years of age (13/17; 65%) (Fig. 11). When the tracheostomy was performed in patients younger than 1 year of age, decannulation was less likely than when it was performed in older children (11/32; 34%) vs. 5/9; 56%), although the difference was not significant ($P = 0.44$) (Fig. 10).

Table 2 Airway obstruction and tracheostomy

	Airway obstruction (+)	Airway obstruction (−)	Unknown	Total
Tracheostomy (+)	40 (93%)*	3	0	43
Tracheostomy (−)	33 (7%)*	440	0	473
Not mentioned	0	0	2	2
Total	73	443	2	518

*Significant difference ($P = 3.41E–54$)

Airway obstruction and tracheostomy (Table 2)

Forty (93%) of the 43 patients required a tracheostomy to relieve airway obstruction, while only 3 underwent a tracheostomy without any obstruction symptoms, which is

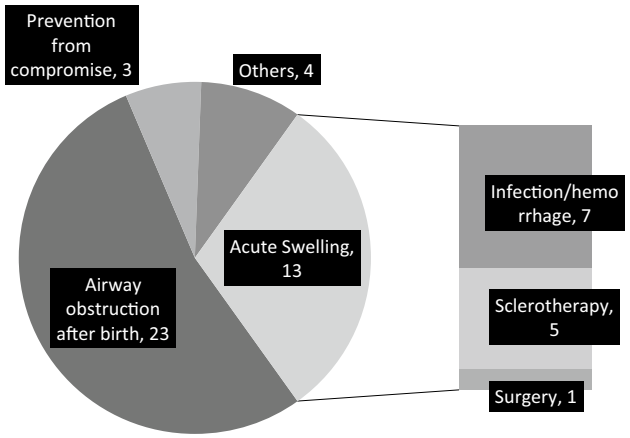


Fig. 12 Direct indications for tracheostomy

Table 3 Direct indications for tracheostomy and decannulation

Reason for tracheostomy	Decannulation (−)	Decannulation (+)	Total
Airway obstruction after birth	17	6	23
Acute swelling	6	7	13
Infection/hemorrhage	2	5	7
Sclerotherapy	3	2	5
Surgery	1	0	1
Prevention from compromise	1	2	3
Others	2	2	4
Total	26	17	43

significant ($P = 3.41E{-}54$). Notably, 33 of 73 patients who had experienced airway obstruction were managed without tracheostomy (Table 2).

Causes and decannulation (Fig. 12; Table 3)

Direct indications for tracheostomy included airway obstruction symptoms in 23 patients, acute swelling from infection and/or hemorrhage in 7, sclerotherapy in 5 and surgery in 1. In three patients, tracheostomy was performed to prevent respiratory compromise before therapy (Fig. 12). In 17 patients whose tracheostomy was closed, airway obstruction after birth was the reason for tracheostomy in 6, acute swelling in 7, and as a preventive strategy in 2. There may be some differences between the reasons for temporary and non-temporary tracheostomy, but those were not significant (Table 3).

Tracheostomy and airway contact of the lesion

The questionnaire asked whether the lesion was in contact with the airway at six anatomical levels based on radiological images: the upper pharynx, middle pharynx (above the epiglottis), larynx above the vocal cord, larynx below the vocal cord, cervical trachea and intrathoracic trachea (Fig. 4). It also asked about the degree to which the lesion circumscribed the area of airway it was in contact with: less than a quarter, a quarter to half, half to three-quarters, three-quarters to total, and totally circumferential.

Significance of airway contact (Table 4)

The lesion was in contact with the airway in 32 (74%) of the 43 patients who required tracheostomy, but in only 58 (12%) of the 473 who were treated without tracheostomy. This demonstrates that patients with a lesion in contact with the airway are significantly more likely to need tracheostomy. However, the lesions in six patients who required a tracheostomy were described as "not having airway contact". The reasons for tracheostomy in these patients were airway obstruction in four, (after birth in two and acute swelling caused by infection in two), as a preventative measure before treatment in one, and for long-term care in one (Table 4).

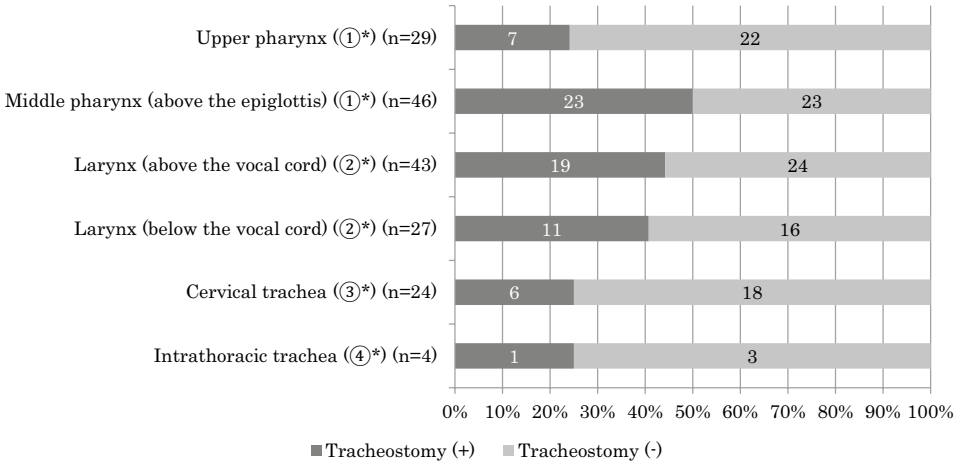
Anatomical site(s) of contact (Fig. 13)

There were 90 patients with a lesion in contact with the airway. The six anatomical levels of contact mentioned above are also summarized in terms of tracheostomy requirement in Fig. 13. The major contact areas were the middle pharynx and larynx above the vocal cord. No specific contact area was significantly associated with the necessity of tracheostomy, although 40% of patients with a lesion in contact with the middle pharynx and larynx underwent tracheostomy (Fig. 13). When comparing the longitudinal extent of the

Table 4 Tracheostomy and airway contact

	Airway contact (+)	Airway contact (−)	Airway contact unknown	Total
Tracheostomy (+)	32 (74%)*	6(14%)	5	43
Tracheostomy (−)	58 (12%)*	398(84%)	17	473
Not mentioned	0	0	2	2
Total	90	404	24	518

*Significant difference ($P = 5.37E{-}28$)



①*—④* indicate the anatomical areas in Fig. 4

Fig. 13 Tracheostomy and airway contact

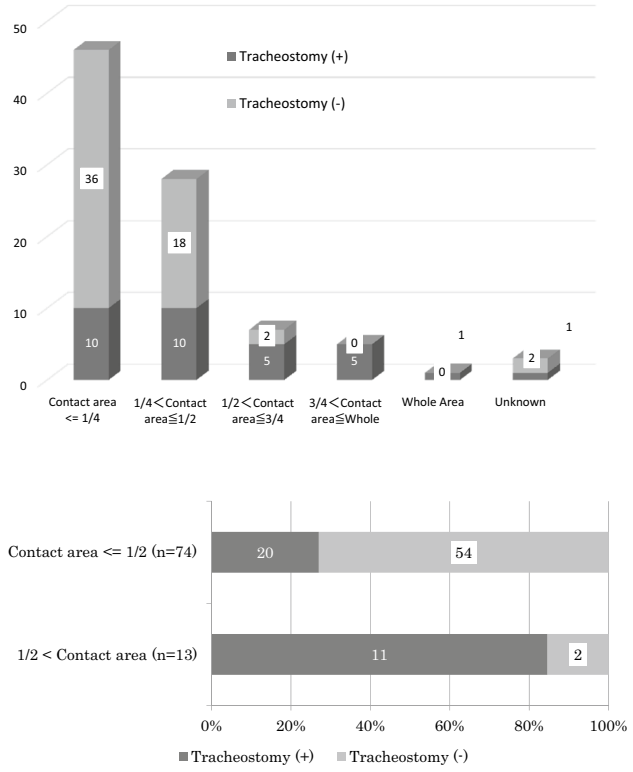
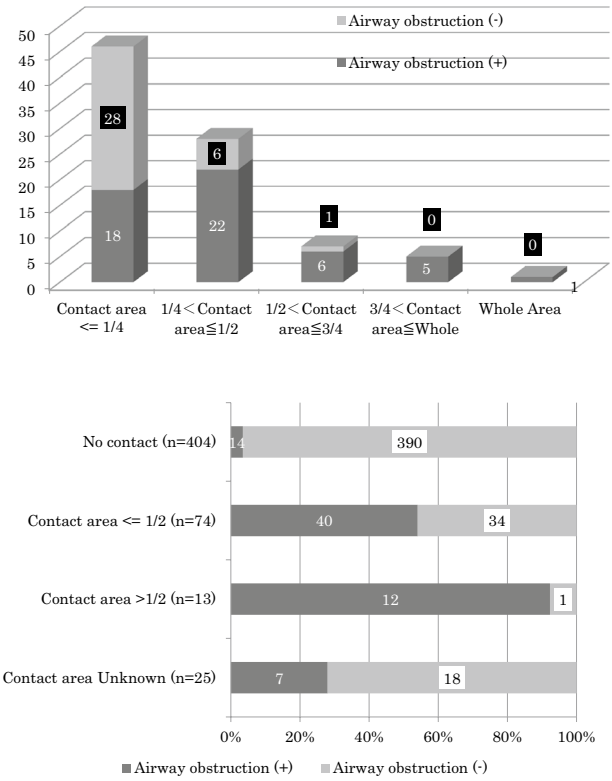


Fig. 14 1, 2 Tracheostomy and range of airway contact



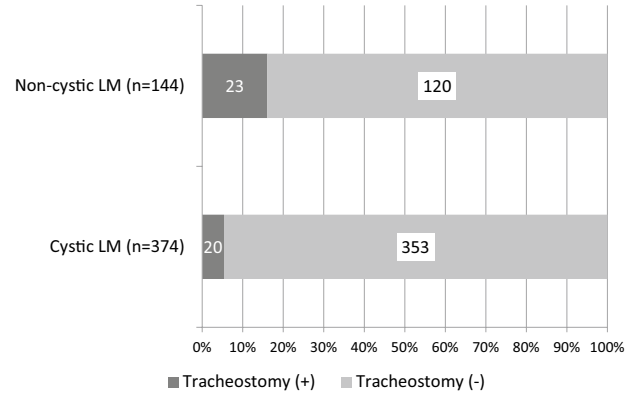
* indicates significant difference (P=0.012)

Fig. 15 1, 2 Airway obstruction and range of airway contact

airway contact of the lesion based on the need for tracheostomy, the longer the anatomical level involved, the higher the rate of tracheostomy requirement, although not to a significant degree (data not shown).

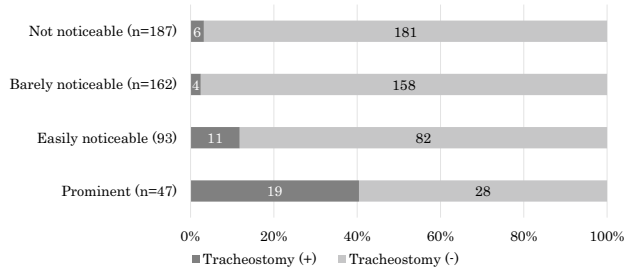
Circumferential area of contact (Figs. 14, 15)

On the other hand, when the circumferential area of contact of the lesion at each anatomical level was compared, a tracheostomy was required significantly more often if the lesion contacted the airway more circumferentially ($P = 0.0001$). In fact, 11 of 13 patients with an LM lesion surrounding



* indicates significant difference (P=8.01E-05)
LM, lymphatic malformation

Fig. 16 Tracheostomy and lymphatic malformation pathology



* indicates significant difference (P=9.85E-05)

Fig. 17 Tracheostomy and disfigurement

more than half of a certain area of the airway underwent tracheostomy (Fig. 14-1, -2) (Table 5). Significantly more patients with a lesion in contact with the airway circumferentially suffered airway obstruction ($P = 0.012$) (Fig. 15-1, -2) (Table 6).

Tracheostomy and other factors (Figs. 16, 17)

We also evaluated the association of tracheostomy with pathological classification as well as with disfigurement. Tracheostomy was associated significantly more with the pathology of non-cystic LM than cystic LM (Fig. 16). LM with more disfigurement was also significantly associated with tracheostomy (Fig. 17).

Discussion

The prevalence of LM is reportedly 1–3 per 10,000 live births. It is most commonly found in the head and neck and sometimes extends to the mediastinum [1]. In the most recent large series of patients with LM from Norway, the head and neck was the most common site of presentation [13]. Our nationwide survey in Japan allowed us to accumulate 606 pediatric cases of head and neck LM, with or without mediastinal involvement. The 87 cases of LM with mediastinal involvement are discussed in another paper [12]. In the present study, we analyzed 518 pediatric patients with head and neck lesions, but without mediastinal LM, accounting for 30.2% (518/1718) of all LM cases registered in our survey, supporting the fact that head and neck is the most common site.

Superficial head and neck LM can be diagnosed in utero or soon after birth. Surveyed data showed that 258 of the 518 (50%) cases in Japan were diagnosed within the neonatal period, including 112 cases detected in utero (Fig. 6). Since the lesion is congenital, it should be apparent at birth, but the survey suggested that LM may be noticed later in life. Since the lesion was described as "not apparent" or "barely noticeable" in 350 (68%) of the 518 patients surveyed (Fig. 8), LM may grow during early life, or caregivers may notice a subtle change. In a recent series of 40 patients with cervicofacial LM reported by Cho et al. from Korea, 73% presented with symptoms before the age of 2 years [14].

Head and neck LM can be large enough to occupy the thoracic outlet and compress the airway and it may be susceptible to infection, which can cause a variety of respiratory symptoms triggered by airway obstruction. When a patient with LM suffers severe obstruction, airway management is crucial to relieve the symptom. Teresa et al. reviewed 141 patients with LM of the upper aerodigestive tract and reported that a high percentage had LM involvement of the oral cavity and oropharynx, but that there was no involvement of the glottis, subglottis, or trachea. They concluded that airway involvement of head and neck LM may occur at multiple sites above the glottis [5]. Berg et al. reviewed 16 patients with laryngeal LM and classified them by a 4-part staging system. They reported that patients with more advanced stage disease required a tracheostomy [15].

Pediatric patients with upper airway obstructions represented by subglottic stenosis and vocal cord palsy may require a tracheostomy, as may those with long-standing compromising respiratory symptoms. Cystic hygroma, or head and neck LM, are uncommon and require tracheostomy [4, 6]. However, the airway management of early infants with head and neck LM is critical for sustaining life, especially when the LM is diagnosed prenatally [16]. *Ex Utero* Intrapartum Therapy (EXIT) has been introduced for prenatally diagnosed LM that may cause airway obstruction soon after birth, and LM is an indication for EXIT [17–19]. Maintaining the airway during EXIT includes manual bagging and/or endotracheal intubation and a tracheostomy is the final option [18]. However, tracheostomy results in speechless life and undermines quality of life. It has been reported

that tracheostomy has wide-ranging effects on quality of life, including the speech and language development of both the patient and their caregivers [20]. Crucial factors are the age at tracheostomy and the time until decannulation. Achieving the earliest possible decannulation could improve the chance of normal speech and language development [10, 11].

Although head and neck LM can be life threatening by compromising the airway, considering that the lesion is generally benign and is sustained later in life, avoiding unnecessary tracheostomy and encouraging the psycho-social development of the patient by speech should be mandatory. For that purpose, there should be some valid indicator(s) for tracheostomy for the LM, but no such index has been published. Thus, the clinical features of patients with or without tracheostomy according to the survey were analyzed to elucidate the conditions that require tracheostomy.

Our survey revealed that most cases of head and neck LM did not require tracheostomy (43/518; 8.3%) (Table 2). Moreover, most tracheostomies were performed in patients younger than 1 year of age (32/43; 74%) because of airway obstruction, but it was closed in 40% of them (17/43), usually when the patient was over 1 year of age (15/17) (Table 3; Figs. 10, 11). Airway obstruction after birth was the leading indication for tracheostomy, which was often permanent for patients with symptoms (Fig. 12). Since it is indispensable to establish the indication for tracheostomy early in life to secure the airway, accumulated data were analyzed to elucidate the conditions associated with tracheostomy.

The questionnaire did not contain any word suggesting "near miss syndrome" or when airway obstruction developed, except soon after birth. However, answers about airway obstruction should reflect the episode of the syndrome. Almost all patients who required a tracheostomy had suffered airway obstruction but 33 of 73 patients who had experienced airway obstruction during their clinical course were managed without tracheostomy (Table 2).

Evaluation of the age distribution of tracheostomy and decannulation revealed that obstructive episodes were most likely to occur within a year of birth, with subsequent decannulation for selected patients (Figs. 10, 11). The reasons for temporary and non-temporary tracheostomy differ somewhat, although not significantly. Shrinkage of cystic LM after treatment allowed for decannulation in 7 of 13 patients who underwent tracheostomy for acute swelling caused by infection/hemorrhage or were treated by sclerotherapy (Table 3). These data suggest that airway obstruction symptoms do not necessarily require tracheostomy if they are temporary or not severe. Ultimately, while every effort is made to avoid unnecessary tracheostomy, which can have negative influence on development, a tracheostomy must be performed in time before airway obstruction causes brain damage. The best timing to perform tracheostomy was not defined by this retrospective survey, but analyzing it would provide clues as to when to consider tracheostomy.

The lesion was in contact with the airway to some degree in 84% (32/43) of the patients with tracheostomy, but in only 12% (58/473) of those without tracheostomy (Table 4). Furthermore, the more the lesion surrounds the airway, the greater the chance that a tracheostomy will be needed, especially when more than half of an area is circumscribed (Table 5). Non-cystic LM and severe disfigurement by the lesion are significantly associated with tracheostomy. Moreover, airway contact, pathology, and disfigurement are all thought to be closely related to each other. The analysis in this paper is mono-variant and a multi-variant analysis should be considered to clarify whether each factor is independent.

To predict the need for tracheostomy for head and neck LM, it may be useful to monitor its contact with the airway radiologically using ultrasonography and/or magnetic resonant imaging (MRI). When the lesion is in contact circumferentially, even if there are no airway obstruction symptoms yet, the need for tracheostomy to prevent respiratory distress can be predicted before therapeutic intervention. If this was detected prenatally, it could suggest the need for EXIT to secure the airway and prevent compromised respiration after

Table 6 Airway obstruction and range of airway contact

Maximum contact area	Airway obstruction (+)	Airway obstruction (−)	Airway obstruction unknown	Total
No contact	14	390	0	404
Contact area < = 1/2	40 (54%)*	34 (46%)*	0	74
Contact area > 1/2	12 (92%)*	1 (8%)*	0	13
Contact area unknown	7	18	2	27
Total	73	443	2	518

*Significant difference ($P = 0.012$)

Table 5 Tracheostomy and range of airway contact

Maximum circumferential area	Tracheostomy (+)	Tracheostomy (−)	Total
Contact area < = 1/2	20 (27%)*	54 (93%)*	74
1/2 < contact area	11 (85%)*	2 (3%)*	13
Total	32	58	90

*Significant difference ($P = 0.0001$)

birth. On the other hand, if there is minimal contact with the airway, we should avoid tracheostomy, which can impact development negatively. Even if the EXIT is considered, it would be more appropriate to try to secure the airway with a tracheal tube first. Further study is needed to clarify this.

Conclusions

Analysis of nationwide survey data on head and neck LM without mediastinal involvement in Japan confirmed that LM, which is congenital, can cause life-threatening airway obstruction symptoms in infancy. Tracheostomy was required when the lesion caused airway obstruction. To elucidate the conditions that would result in LM compromising the airway and necessitating tracheostomy, accumulated clinical features were analyzed. Tracheostomy should be considered when the lesion is in contact with the airway and surrounds more than a semicircle; however, if contact with the airway is minimal, we should avoid performing tracheostomy, which impacts negatively on development. Even if the EXIT is considered, it would be more appropriate to try to secure the airway with a tracheal tube first. Further study is indispensable to elucidate the indications.

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Compliance with ethical standards

Conflict of interest We have no conflicts of interest to declare.

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