Long-term Outcome of the Management of Otitis Media with Effusion in Children with and Without Cleft Palate Using the House-brand Polyethylene Ventilation Tube Insertion

Kitirat Ungkanont, M.D.*, Alisa Tabthong, M.D., Chulaluk Komoltri, Ph.D.***, Amornrut Leelaporn, Ph.D.****, Vannipa Vathanophas, M.D.*, Archwin Tanphaichitr, M.D.*, Thidarat Wensanthia******

**Department of Otorhinolaryngology, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok 10700, Thailand, **Uthaithani Hospital, Mueang District, Uthai Thani 61000, Thailand,*

****Office for Research and Development, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok 10700, Thailand, ****Department of Microbiology, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok 10700, Thailand.*

ABSTRACT

Objective: To study the long-term outcome of otitis media with effusion in children with and without cleft palate treated with the same protocol of ventilation tube insertion.

Materials and Methods: A retrospective cohort study was conducted in eighty-five children with cleft palate and 80 children without cleft palate who had otitis media with effusion and had follow-up between 2001 and 2019. Both groups were treated with ventilation tube insertion for longstanding middle ear effusion more than 90 days. The main outcome was the cumulative incidence of surgical management, time of the indwelling ventilation tubes, conditions of the tympanic membrane, and the hearing outcome.

Results: At 24 months old, 63.5% of children with cleft palate and 11.3% of children without cleft palate had their first ventilation tube insertion. Repeated surgery was done in 81.2% of children with cleft palate and 50% of children without cleft palate ($p < 0.001$). The median duration of the indwelling tube was 11.3 months in the children with cleft palate and 12.4 months in the non-cleft children ($p = 0.82$). At the end of the study, 63.7% of children without cleft palate and 43.5% of children with cleft palate had normal tympanic membrane ($p = 0.009$). The hearing outcomes of children with and without cleft palate were 20.7 dB and 19.3 dB, respectively.

Conclusion: Children with and without cleft palate were managed under the same guideline and the hearing outcome was favorable in both groups.

Keywords: Cleft palate; otitis media with effusion; ventilation tube (Siriraj Med J 2021; 73: 245-251)

INTRODUCTION

Cleft palate is an important underlying factor for otitis media in infants and children. The incidence of non-syndromic cleft palate without cleft lip is 1:2,000 live births.[1] Anatomical abnormalities of the eustachian tube in cleft palate include abnormal insertion of the levator veli palatini and tensor veli palatini muscle, a small paratubal muscle, greater cartilage cell density, less elastin in the hinge portion, and a smaller angle between the tensor veli palatini muscle and the eustachian tube.[2,3]

Corresponding author: Kitirat Ungkanont
E-mail: kitirat.ung@mahidol.ac.th
Received 29 December 2020 Revised 8 February 2021 Accepted 11 February 2021
ORCID ID: http://orcid.org/0000-0003-0923-1908
http://dx.doi.org/10.33192/Smj.2021.32

Relocation of the levator veli palatini muscle during palatoplasty has been reported to have no effect on the eustachian tube function.[4] Da Silva et al.[5] found that the middle ear status had no correlation with velopharyngeal function after palatoplasty. Also, the type of cleft palate has been reported to have no association with the number of ventilation tube insertions.[6] Children with cleft palate are more prone to otitis media with effusion (OME) than children without cleft palate. Previous studies reported that children with cleft palate had an onset of OME in infancy with prolonged effusion, higher incidence of complications and hearing loss from chronic OME.[7,8]

Myringotomy and ventilation tube insertion are accepted as the most effective management for chronic middle ear effusion and hearing loss in children with cleft palate. The timing of the ventilation tube insertion and the management protocol in children with cleft palate are the main controversial issues and remain under extensive review.[9,10] Insertion of ventilation tubes in children with cleft palate can be done in a routine fashion or only when chronic effusion is present. There are no data in the literature to support early routine ventilation tube insertion in children with cleft palate in terms of the benefit of the outcome, especially when effusion is not found. From the clinical practice guideline ventilation tube insertion in at-risk children should be done in cases with persistent middle ear effusion longer than 90 days or effusion that is unlikely to resolve itself as demonstrated by a type-B (flat) tympanogram.[11] We present herein the results of a retrospective cohort study of OME in children with and without cleft palate. Both groups were treated with regular follow-up and ventilation tube insertion for longstanding middle ear effusion more than 90 days. The main outcome was the cumulative incidence of OME, age incidence and number of tympanostomy tube insertion, time of the indwelling tubes, age of resolution of OME, outcome of the tympanic membrane (TM), and the hearing outcome.

MATERIALS AND METHODS

This retrospective cohort study was conducted in children with and without cleft palate who had OME and had regular follow-up between January 2001 and May 2019. The research was approved by the institutional review board of our institute. Children with cleft palate or craniofacial anomalies were first seen by the plastic surgery service and they were referred to the Pediatric Otolaryngology clinic for ear evaluation. Children without cleft palate came to the Pediatric Otolaryngology clinic for ear problems, hearing problems, or for an evaluation of their speech and development. Children with congenital syndromic and non-syndromic sensorineural hearing loss were excluded from the study. Data gathering included the demographic data, physical examination, diagnosis, and hearing test results. Diagnosis of OME was done by pneumatic otoscopy. Pneumatic oto-endoscopy was done in some cases with difficult diagnosis to add extra visualization of the TM and the middle ear. All ear examinations were done in the outpatient department. Ear events during the follow-up period were recorded. The definition of acute otitis media (AOM) was an acute inflammation of the middle ear, with bulging of the TM and middle ear effusion. Otitis media with effusion (OME) was defined as middle ear effusion without symptoms and signs of acute inflammation. A hearing test was done as early as possible after the first visit. In young infants, Auditory brain stem response (ABR) and auditory steady state response (ASSR) tests were done. Hearing tests were done periodically according to the ear events and after each surgery. Pure tone audiometry was done when the child was old enough to be able to co-operate.

The indication of myringotomy with ventilation tube insertion was the presence of longstanding middle ear fluid for more than 90 days with or without hearing loss. Type-B tympanogram was not included in the routine investigations because tympanometry might not be reliable in very young children. If chronic middle ear fluid was found near the date of the scheduled palatoplasty, the first myringotomy and ventilation tube insertion were done in the same setting. After surgery, the patients had regular follow-up every 8–12 weeks until their ear conditions were resolved. In patients with ventilation tubes, the condition of the TM and ventilation tubes were recorded until the tubes fell out. The duration time of the indwelling tubes were recorded.

Middle ear fluid culture was done in patients who had myringotomy and ventilation tube insertion. Middle ear fluid samples were aseptically collected and sent to the microbiology laboratory within 30 minutes for Gram staining and culture. The specimen was inoculated onto chocolate agar, 5% sheep blood agar, and MacConkey agar plates for the cultivation of aerobic and facultative anaerobic bacteria and were then examined after 24, 48, and 72 hours of incubation. The results of the middle ear fluid culture were recorded.

All children with cleft palate were scheduled for regular follow-up until no more recurrence of OME was found. Children without cleft palate were re-evaluated for their ear events at intervals until the middle ear effusion or abnormalities of the TM were no longer present. The outcome of the treatment was recorded at the last follow-up by ear examination and hearing test.

A favorable outcome was defined as a normal TM with no middle ear effusion and a normal hearing threshold less than or equal to 20 decibels (dB).

Descriptive statistics were used for the demographic and other descriptive data. Univariate analysis was done for comparisons between the data of the children with and without cleft palate using chi-square, T-test, or non-parametric statistics. The cumulative incidence of first ventilation tube insertion for OME and the resolution of OME with a normal TM were plotted between the events and the age of the patients. A Kaplan–Meier life table plot was used for the duration of the indwelling ventilation tubes. Statistical analysis was done by using SPSS version 22.

RESULTS

The data of 165 pediatric patients with OME were included in the analysis, comprising 85 children with cleft palate and 80 children without cleft palate. The children's demographic data are shown in Table 1. The children with cleft palate had an associated cleft lip in 48 cases (56.5%). There was a significant difference in age at first contact for the children with and children without cleft palate. Most of the children with cleft palate (72.9%) were referred to the Pediatric Otolaryngology clinic when they were younger than 24 months old, while children without cleft palate were seen in the clinic at the median age of 4 years old.

Ear events in this series consisted of acute otitis media (AOM), otitis media with effusion (OME), and otorrhea through ventilation tube. The rate of AOM was 1.96±5.1 per year in children with cleft palate and 1±3.2 per year in children without cleft palate ($p = 0.147$). The rate of otorrhea in children with cleft palate was 7.2 per year and 0.73 per year in children without cleft palate ($p = 0.003$). There was no cholesteatoma and no other suppurative complications of otitis media in this series.

All of the patients in this series had chronic OME requiring myringotomy and ventilation tube insertion. The cumulative percentage of patients who had first myringotomy and ventilation tube insertion according to age is shown in Fig 1. The cumulative incidence of OME that met the criteria for myringotomy and ventilation tube insertion was significantly higher in children with cleft palate, especially in the first 5 years of life. At 24 months old, 63.5% of the children with cleft palate had their first myringotomy compared to 11.3% of the children without cleft palate. Most of the children without cleft palate had their first myringotomy between 24 to 72 months old. Repeated ventilation tube insertion was done in 69 cases (81.2%) of children with cleft palate and in 40 cases (50%) of children without cleft palate ($p < 0.001$). The mean number of myringotomy and ventilation tube insertions throughout the cohort was 2.7 in the children with cleft palate and 1.9 in the children without cleft palate ($p = 0.002$).

TABLE 1. Demographic data of the children with and without cleft palate.

	Cleft	Non-cleft	P-value
Number of patients	**85**	**80**	
Gender			
Male	48 (56.5%)	44 (55%)	0.85
Female	37 (43.5%)	36 (45%)	
Age (months)			
first contact	8 (25)*	49.5 (55)*	< 0.001**
first myringotomy	17 (50)*	51 (51)*	< 0.001**
last contact	95.5±62.3	115.8±57.2	0.03
Follow-up duration (months)	73.7±49.3	57.8±38.8	0.02

*median and interquartile range.

** non-parametric statistics.

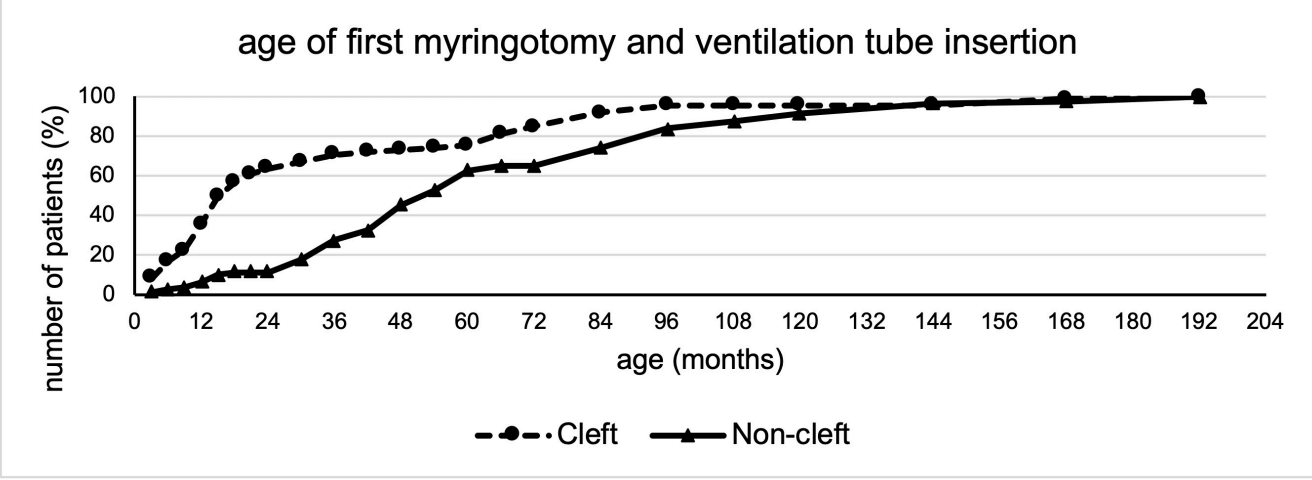


Fig 1. Cumulative percentage of patients who had first myringotomy and ventilation tube insertion for OME according to age
OME = otitis media with effusion.

The duration of the indwelling ventilation tubes was compared between 72 children with cleft palate and 62 children without cleft palate who used a house-brand ventilation tube, which was made from a polyethylene tube and shaped like a bobbin. Our study cohort included patients who used other tubes, such as the modified Armstrong tube, Shepard tube, T-tube, and Shah tube, but the sample size was too small for comparison. The duration times of the indwelling tubes in children with and without cleft palate are shown in Fig 2. The median duration time of the indwelling tube was 11.3 months (95% confidence interval = 9.2,13.4) in the children with cleft palate and 12.4 months (95% confidence interval = 9.5,15.3) in the children without cleft palate (p = 0.82). The mean total follow-up time until resolution of OME was 73.7±49.3 months in cleft children and 57.8 ± 38.8 months in non-cleft children.

Bacterial culture of the middle ear fluid from the first myringotomy was done. From the specimens of the 165 patients, 156 bacterial isolates were found. No growth was found in 76 cases. The prevalence of bacteria in the middle ear fluid was similar in the children with and without cleft palate. Coagulase-negative Staphylococcus was the most common bacteria found in both groups, followed by *Staphylococcus aureus* and *Haemophilus influenzae*.

The outcomes of the management of OME are shown in Table 2. The children without cleft palate had more intact TM than the children with cleft palate with statistical significance (p = 0.009). The mean age at the last contact with a bilateral intact TM was 98.7 months old in the children with cleft palate and 118.4 months old in the children without cleft palate (p = 0.14). The rate of TM perforation was not different between the children with cleft palate and the children without cleft palate.

The hearing threshold at the first contact and subsequent hearing evaluation were obtained by ABR, ASSR, speech reception threshold or pure tone audiometry according to the patient's age and co-operation. Visual reinforcement audiometry (VRA), otoacoustic emission and tympanogram were not done routinely but they were used in selected cases. The hearing threshold was shown in Table 2. At the end of the study, 66.2% of the children with cleft palate and 69.6% of the children without cleft palate had normal hearing. The average hearing threshold was 20.7 dB in cleft children and 19.3 dB in non-cleft children.

The cumulative rate of resolution of OME in children with cleft palate is shown in Fig 3. The earliest resolution of OME was found in one case at 3 years old, but the peak incidence of resolution occurred between 5 to 8 years old. At 10 years old, the resolution rate of OME in the children with cleft palate was 83.8%. Fifty-one percent of the children with cleft palate had normal hearing at 7 years old and 80.4% of the children had normal hearing at 10 years old.

DISCUSSION

Children with cleft palate are identified as "at-risk" children for OME.[12] Hearing loss from OME may add more severity to any speech defect that already exists from cleft palate. In our previous study,[13] we found that the cumulative incidence of OME in children with cleft palate was 53.7% at 12 months of age and 81.1% when the children reached 24 months old. Chronic middle

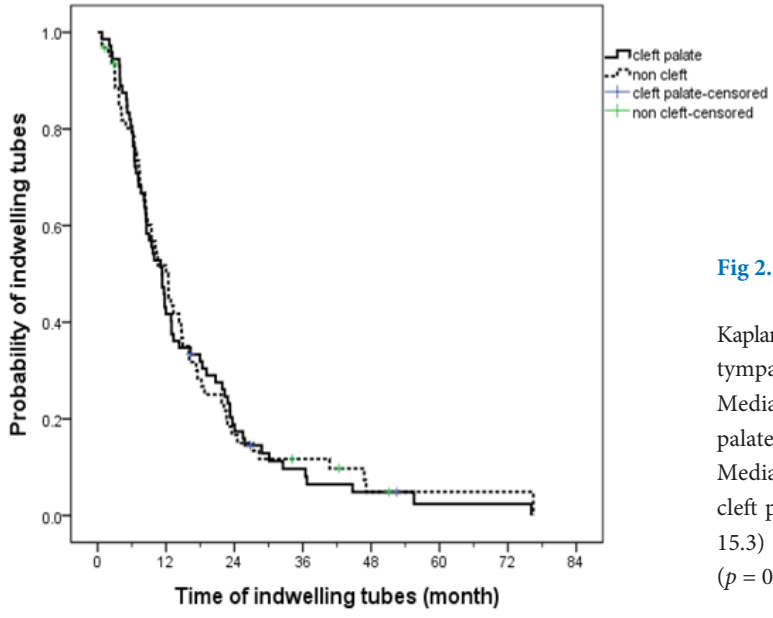


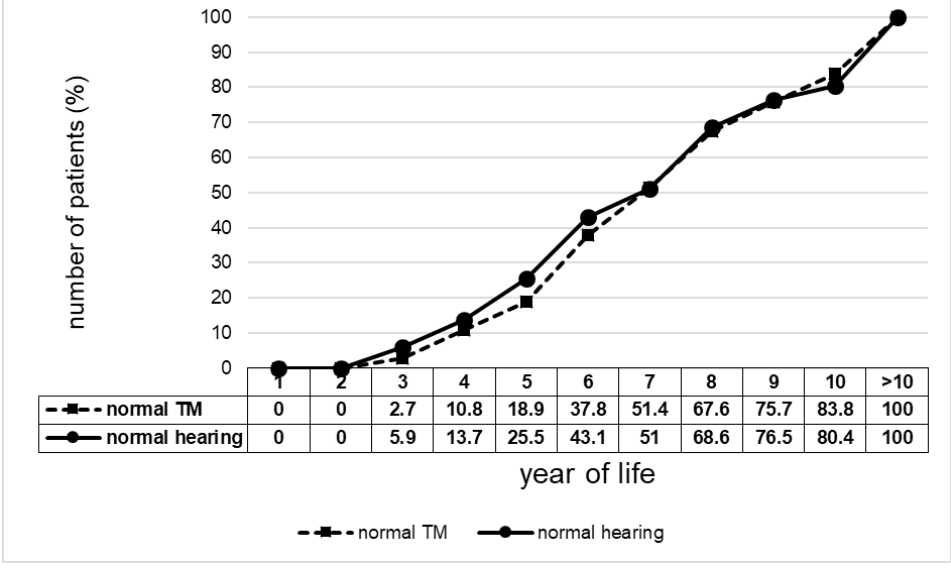
Fig 2. Time in place of the indwelling tubes

Kaplan–Meier curves showing the time in place of the indwelling tympanostomy tube
Median time of the indwelling tube in the children with cleft palate = 11.3 months (95% confidence interval = 9.2, 13.4)
Median time of the indwelling tube in the children without cleft palate = 12.4 months (95% confidence interval = 9.5, 15.3)
(p = 0.82)

TABLE 2. Outcome of the management of otitis media with effusion in the children with and without cleft palate.

	Cleft	Non-cleft	P-value
TM and middle ear			
Intact (bilateral)	37/85 (43.5%)	51/80 (63.7%)	0.009*
Perforation (at least one side)	9/85 (10.6%)	5/80 (6.2%)	0.318
Tube in place (at least one side)	31/85 (36.5%)	14/80 (17.5%)	0.006*
Residual effusion (at least one side)	8/85 (9.4%)	9/80 (11.2%)	0.698
Residual retraction	0	1 (1.2%)	-
Total	85	80	
Hearing threshold (dB)			
First hearing	40.2	42.6	0.40
Last hearing	20.7	19.3	0.52

TM = tympanic membrane.



	1	2	3	4	5	6	7	8	9	10	>10
normal TM	0	0	2.7	10.8	18.9	37.8	51.4	67.6	75.7	83.8	100
normal hearing	0	0	5.9	13.7	25.5	43.1	51	68.6	76.5	80.4	100

Fig 3. Cumulative percentage of the children with cleft palate who had a resolution of OME with a normal tympanic membrane/ normal hearing according to age
OME = otitis media with effusion.

ear effusion and frequent recurrences are characteristic of OME in children with cleft palate. Ear and hearing evaluation should be done as early as possible because OME in children with cleft palate usually presents when they are young infants. Regular ear examination should be done to detect the recurrence or longstanding middle ear effusion justifying ventilation tube insertion. There is no clinical evidence to support early prophylactic ventilation tubes in terms of speech and language development.[9] Smillie et al.[14] concluded from their study that the same guideline should be used for ventilation tube insertion in children with and without cleft palate as the complication rate was not higher in children with cleft palate.

Children with cleft palate had their first myringotomy at a younger age when compared to children without cleft palate. In the systematic review by Felton et al[15], there were some studies that children with cleft palate who had early placement of tympanostomy tubes at the time of cleft lip repair (at 3-4 months old) had achieved normal hearing and speech comparable to the non-cleft children. However, this group of infants had higher rate of otorrhea than older children.[15] Most of the children with cleft palate in our study had their tympanostomy tube insertion at the time of palatoplasty. In our study, 70% of the children with cleft palate had their first myringotomy before they were 3 years old, while 62.5% of the children without cleft palate had their first ventilation tubes within their first 5 years. From the study of Huang et al.,[16] the mean age at first myringotomy in children with cleft palate was 1.3 years old, which was about the same age as in our study. The age of the first myringotomy had no effect on the long-term hearing outcome.[17]

The median time of the indwelling tubes remaining in the children with cleft palate was 11.3 months, which was a little shorter than in the children without cleft palate, but without significant difference. The indwelling duration of our house-brand ventilation tube was the same as for the commercial short-term tube, which was 10-18 months. Ahn et al.[18] found that repeated insertion was associated with a shorter indwelling time of the tube.

Middle ear effusion causes mild to moderate hearing loss. In our study, the mean hearing threshold in the children with and without cleft palate with OME before ventilation tube insertion was around 40 dB. The first hearing test in young infants was done by auditory brain stem response (ABR) and auditory steady state response (ASSR). Subsequent pure tone audiometry confirmed that the hearing loss was conductive. The children without cleft palate in this study were older than the children with cleft palate when they entered the study and some of their first hearing tests were done by pure tone audiometry. Sundman et al.[19] reported an ABR threshold of 41.4 dB in infants with OME and 40.1 dB in infants with cleft lip and cleft palate. The median neonatal ABR threshold in infants with cleft palate in the study of Tengroth et al.[20] was 35 dB (range, 20-45 dB). Kim et al.[17] studied the rate of hearing loss and TM perforation in children with cleft palate at 10 years old and found that the rate of hearing loss (>25 dB) was 38.8%. In this study, the mean hearing threshold after the management was normal in both groups without significant difference.

The detection of OME requires regular ear examination as the disease is asymptomatic. Children with OME who are not at risk require regular follow-up every 3-6 months until the fluid disappears to prevent complications, such as retraction or cholesteatoma.[12] At the end of this study, the number of children with cleft palate who still had ventilation tubes in place was greater than in the non-cleft group, so the children with cleft palate would be expected to be followed for a longer period. Regular follow-up with hearing evaluation in children with cleft palate should be continued until the resolution of OME is found and the hearing is stabilized.

The main limitation of this study was related to the retrospective nature of the data. Also, the children without cleft palate could not be considered as a true control because they did not undergo the same surveillance as the children with cleft palate, rather they came in as patients with ear or hearing-related problems. We did not compare the resolution rate of OME and the time to normal hearing between the children with and without cleft palate because the results could not be generalized. Finally, the choice of tympanostomy tube was made according to the patient's reimbursement scheme and the surgeon's preference.

On the other hand, information on the children with cleft palate was collected from the first day that they entered the craniofacial clinic and were placed under surveillance for ear diseases throughout the study. The results of this study should be useful as a description of OME in children with cleft palate in the aspect of the long-term outcome. From this study, a strategy involving careful observation and ventilation tube insertion when the middle ear effusion did not resolve in due course could be useful for children with cleft palate as well as for children without cleft palate. The outcome was comparable in terms of the children achieving a normal hearing threshold, the low rate of TM perforation and no severe complications.

CONCLUSION

The children with cleft palate had their first ventilation tube insertion at a younger age than the children without cleft palate. The children with cleft palate had a greater number of ventilation tubes and more episodes of otorrhea. The duration time of the indwelling tubes was not different in both groups. At the last follow-up, there were more children with cleft palate who still had a ventilation tube in place. Children without cleft palate had a greater number of normal TM than the cleft palate group. The rate of TM perforation was lower in the children without cleft palate, but with no statistical significance. The children with and without cleft palate who had OME were managed with the same guideline and the hearing outcome was favorable in both groups. The children with cleft palate were expected to have a longer period of follow-up.

ACKNOWLEDGEMENTS

The authors would like to acknowledge Miss Jeerapa Kerdnoppakhun, Miss Ngamrat Treerassapanich, and Miss Chutima Polsrimueng for their help in gathering the data and preparing the manuscript.

Conflict of Interest: The authors declare they have no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding: The authors received no financial support for the research, authorship, and/or publication of this article.

REFERENCES

1. Deleyiannis FWB, Maguire RC. Congenital malformations of the mouth and pharynx: orofacial cleft and related syndromes. In: Bluestone CD, Healy GB, Simons JP, editors. Pediatric Otolaryngology. 2. 5th ed. Shelton: People's Medical Publishing House-USA; 2014.p.1879-84.
2. Takasaki K, Sando I, Balaban CD, Ishijima K. Postnatal development of eustachian tube cartilage. A study of normal and cleft palate cases. Int J Pediatr Otorhinolaryngol 2000;52: 31-6.
3. Heidsieck DS, Smarius BJ, Oomen KP, Breugem CC. The role of the tensor veli palatini muscle in the development of cleft palate-associated middle ear problems. Clin Oral Investig 2016; 20:1389-401.
4. Kobayashi H, Sakuma T, Terao H, Sanbe T, Suzaki H, Ohkubo F. Palatoplasty and growth of the temporal bone mastoid air cells. Cleft Palate Craniofac J 2012;49:541-5.
5. da Silva DP, Collares MV, da Costa SS. Effects of velopharyngeal dysfunction on middle ear of repaired cleft palate patients. Cleft Palate Craniofac J 2010;47:225-33.
6. Garcia-Vaquero C, Mir C, Graterol D, Ortiz N, Rochera-Villach MI, ME LL, et al. Otologic, audiometric and speech findings in patients undergoing surgery for cleft palate. BMC Pediatr 2018;18:350.
7. Kobayashi H, Sakuma T, Yamada N, Suzaki H. Clinical outcomes of ventilation tube placement in children with cleft palate. Int J Pediatr Otorhinolaryngol 2012;76:718-21.
8. Skinner ML, Tunkel DE. Primary care of infants and children with cleft palate. In: Bluestone CD, Healy GB, Simons JP, editors. Pediatric Otolaryngology. 2. 5th ed. Shelton: People's Medical Publishing House-USA; 2014.p.1879-84.
9. Ponduri S, Bradley R, Ellis PE, Brookes ST, Sandy JR, Ness AR. The management of otitis media with early routine insertion of grommets in children with cleft palate -- a systematic review. Cleft Palate Craniofac J 2009;46:30-38.
10. Karanth TK, Whittemore KR. Middle-ear disease in children with cleft palate. Auris Nasus Larynx 2018;45:1143-51.
11. Rosenfeld RM, Schwartz SR, Pynnonen MA, Tunkel DE, Hussey HM, Fichera JS, et al. Clinical practice guideline: Tympanostomy tubes in children. Otolaryngol Head Neck Surg 2013;149(1 Suppl):S1-35.
12. Rosenfeld RM, Shin JJ, Schwartz SR, Coggins R, Gagnon L, Hackell JM, et al. Clinical Practice Guideline: Otitis Media with Effusion (Update). Otolaryngol Head Neck Surg 2016;154 (1 Suppl):S1-S41.
13. Ungkanont K, Boonyabut P, Komoltri C, Tanphaichitr A, Vathanophas V. Surveillance of Otitis Media With Effusion in Thai Children With Cleft Palate: Cumulative Incidence and Outcome of the Management. Cleft Palate Craniofac J 2018;55: 590-5.
14. Smillie I, Robertson S, Yule A, Wynne DM, Russell CJ. Complications of ventilation tube insertion in children with and without cleft palate: a nested case-control comparison. JAMA Otolaryngol Head Neck Surg 2014;140:940-3.
15. Felton M, Lee JW, Balumuka DD, Arneja JS, Chadha NK. Early Placement of Ventilation Tubes in Infants with Cleft Lip and Palate: A Systematic Review. Otolaryngol Head Neck Surg 2018;158:459-64.
16. Huang CY, Wu CS, Tang CH, Wang MC, Kuo TY, Tu TY. Palatoplasty decreases the re-insertion rate of middle ear ventilation tube in cleft palate children - A population-based birth cohort study. Acta Otolaryngol 2016;136:768-74.
17. Kim E, Kanack MD, Dang-Vu MD, Carvalho D, Jones MC, Gosman AA. Evaluation of Ventilation Tube Placement and Long-term Audiologic Outcome in Children With Cleft Palate. Cleft Palate Craniofac J 2017;54:650-5.
18. Ahn JH, Kang WS, Kim JH, Koh KS, Yoon TH. Clinical manifestation and risk factors of children with cleft palate receiving repeated ventilating tube insertions for treatment of recurrent otitis media with effusion. Acta Otolaryngol 2012;132: 702-7.
19. Sundman H, Flynn T, Tengroth B, Lohmander A. ABR thresholds in infants born with CLP and OME and infants with OME. Int J Pediatr Otorhinolaryngol 2016;81:21-5.
20. Tengroth B, Lohmander A, Hederstierna C. Hearing Thresholds in Young Children With Otitis Media With Effusion With and Without Cleft Palate. Cleft Palate Craniofac J 2020;57:616-23.