

OK-432 treatment in pediatric patients with recurrent thyroglossal duct cyst after surgery

Nobuo Ohta¹, Shigeru Fukase², Miho Nakazumi¹, Teruyuki Sato¹, Takahiro Suzuki¹

¹Division of Otolaryngology, Tohoku Medical and Pharmaceutical University Hospital, Sendai, Japan

²Fukase Clinic, Yamagata, Japan

Article history: Received: 20.05.2021 Accepted: 01.06.2021 Published: 16.06.2021

SUMMARY:

Introduction: Recurrent thyroglossal duct cyst after surgery is not a rare condition and first-line treatment has not been established yet.

Aim: Evaluation of outcomes and complications of OK-432 treatment in patients with recurrent thyroglossal duct cyst after surgery.

Material and methods: This study is designed as a case series with planned data collection at Tohoku Medical and Pharmaceutical University and Fukase Clinic. Five patients with recurrent thyroglossal duct cyst after surgery received this therapy between January 2014 and February 2020 on an outpatient basis, without hospitalization. OK-432 solution was injected into the lesion using an 18- or 27-gauge needle, depending on the location and size of the lesion, as well as on possible complications.

Results: Lesions showed marked reduction or total shrinkage in all patients, with no local scarring or deformity at the injection site. Side effects manifested as local pain at the site of injection and fever (37.5–38.5°C) observed in three patients, but the symptoms resolved within a few days.

Conclusions: Since OK-432 therapy is simple, easy, safe and effective, it can be used as an alternative to surgery in the treatment of recurrent thyroglossal duct cyst after surgery.

KEYWORDS:

OK-432, recurrent thyroglossal duct cyst, surgery

INTRODUCTION

Thyroglossal duct cyst is the most common congenital neck cyst. Various surgical procedures, including Sistrunk operation, simple cyst excision, endoscopic surgery through breast approach and robotic surgery with facelift approach, have been implemented to treat this condition [1–8]. Although surgical removal is very effective, postoperative complications are common and include wound infection or cyst recurrence [6]. Recently, OK-432 treatment has been reported as an effective approach in treating ranula, lymphatic malformation, auricular hematoma, mucocele of the lower lip and thyroglossal duct cyst [9–15]. We investigated its effectiveness in patients with recurrent thyroglossal duct cyst after surgery.

MATERIALS AND METHODS

Five pediatric patients with recurrent thyroglossal duct cyst after surgery received OK-432 therapy. Four of these patients were referred to our hospital from other institutions. All patients had previously undergone a Sistrunk operation and all were treated with OK-432 on an outpatient basis, without hospitalization. One representative case is presented here.

OK-432 TREATMENT

In each patient, cyst fluid was aspirated to a maximum degree, with cyst compression applied as needed. The fluid was examined for pathologies. After we had determined the volume of each cyst, a sufficient amount of OK-432 was prepared (Picibanil, Chugai Pharmaceutical Co., Tokyo, Japan) and diluted with saline solution (1 Klinische Einheit [KE]/mL = 0.1 mg/mL). We changed the syringe and injected OK-432 solution into the cyst, using the same needle which had been used for aspiration (the amount of injected OK-432 was equal to about half the volume of previously removed cyst fluid) The injection was successful, as no resistance was noted.

FOLLOW-UP

All patients were regularly checked for an average period of 14.2 months (duration of follow-up period ranged from 12 to 18 months) after the final (first or second) injection. Analgesics on demand were prescribed as prophylaxis for potential fever. Induration and redness of skin at the injection site were observed the day after the injection. Both local and systemic conditions of all patients were evaluated on days 7 and 42 after the injection.

Tab. I. Number and type of surgeries performed (Snow et al. in Tweedie and Jacobs modification).

CASE	AGE (Y)	SEX	SIZE (CM)	NO OF TX	DOSE (KE)	TOTAL DOSE (KE)	FOLLOW-UP (MONTHS)	OUTCOME	PREVIOUS TX	FURTHER TX
1	5	M	3,7	1	1	1	12	TS	Sistrunk	none
2	4	F	4,5	1	1	1	15	TS	Sistrunk	none
3	5	M	2,9	1	1	1	14	TS	Sistrunk	none
4	6	M	2,6	1	1	1	12	TS	Sistrunk	none
5	7	F	3,1	2	1	2	18	TS	Sistrunk	none

Size, maximum diameter (cm); TS, total shrinkage; MR, marked reduction; Follow-up is in months; No., number; Tx, treatments; Further Tx, Further treatment; Previous Tx, Previous treatment; Sistrunk, Sistrunk operation, dose is in KE (Klinische Einheit); total dose is in KE; HT

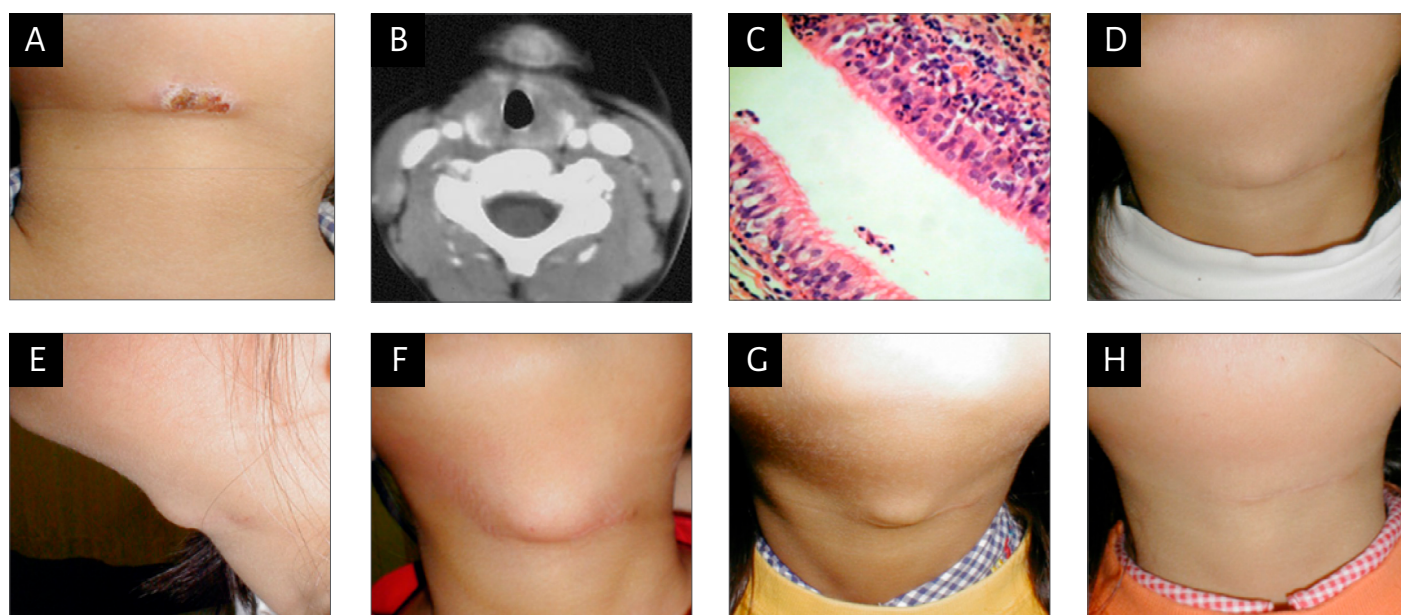


Fig. 1. Total shrinkage of cervical swelling in a 5-year-old Japanese boy after a single dose of treatment with OK-432. (A) Local findings before OK-432 treatment showing a thyroglossal duct cyst of the anterior cervical region with crust (about 3.7 cm). (B) Initial axial computed tomography scan performed before surgery, showing a thyroglossal duct cyst. (C) Pathological findings in a surgical specimen demonstrating that the cyst is predominantly composed of columnar and squamous epithelial lining with focal concentrations of ciliated respiratory epithelium. (D) Anterior and (E) lateral views of the cyst obtained 2 months after surgery. (F) Local findings 2 days after OK-432 treatment showing swelling and slight redness of the lesion. (G) Local findings 3 weeks after OK-432 treatment showing slight swelling of the lesion. (H) Local findings 6 weeks after OK-432 treatment showing total shrinkage of the lesion.

If response to treatment was insufficient, the same volume of OK-432 was administered in a second injection. We defined “total shrinkage” as a complete absence of cyst, “marked reduction” as a decrease of more than half of the cyst size and “partial reduction” as a decrease of less than half of the pretreatment cyst size, as determined by clinical examination or computed tomography.

RESULTS

Five patients with thyroglossal duct cyst after surgery received OK-432 treatment (Tab. I.). The mean patient age was 5.4 years (ranging from 4 to 7 years). The maximum cyst diameter ranged from 2.6 to 4.5 cm (mean 3.4 cm). Either 1 or 2 courses of treatment were administered (mean value 1.2) and the mean follow-up period was 14.2 months (ranging from 12 to 18 months). The dose of OK-432 given at each treatment course was 1 KE and the total administered dose was 1 or 2 KE (mean 1.2 KE). Six therapeutic procedures were performed on 5 lesions; 4 out of 5 patients

required a single course of treatment. All cases showed total shrinkage or marked reduction (follow-up for >12 months after the final injection without recurrence or further treatment) after 1 or 2 OK-432 treatment doses. A representative case is shown in Fig. 1. – total shrinkage after a single injection.

No serious complications other than moderate-grade fever (37.5 to 38.5 °C) for a few days after injection were recorded in our study. Fever was managed with antipyretics. No patient developed infection or abscess. No evidence of scarring on the skin at the injection site was observed in any patient. This study was approved by the Institutional Review Board.

DISCUSSION

Thyroglossal duct cyst results from incomplete involution of the thyroglossal duct. Although surgery is the treatment of choice, hemorrhage, resection of thyroid cartilage, wound infection

and cyst recurrence are common post-operative complications [16]. Risk factors for recurrence of thyroglossal duct cyst include post-operative infection, thyroid drain placement during surgery and applied surgical methods [6, 16–18]. The most important factor predicting recurrence was the type of surgical resection; the recurrence rate recorded after Sistrunk surgery was 5.3% and 55.6% after simple surgical excision. The “extended” Sistrunk procedure is commonly performed [19]. In this study, all 5 patients underwent Sistrunk surgery. OK-432 treatment was very effective in all cases and no serious complications were observed.

OK-432 treatment does not require special equipment or patient hospitalization and leaves no scar or pigmentation on the skin at the injection site. The advantages of OK-432 treatment over surgical modalities can be summarized as follows: (1) the procedure is brief, (2) no local anesthesia is required, (3) the treatment is painless and (4) secondary infections and hemorrhages are rare [9–15]. These benefits are particularly important for pediatric patients. The mechanism responsible for the effectiveness of OK-432 therapy involves intense production of IL-6, IL-8, IFN- γ ,

TNF- α , VEGF and periostin, which are found in fluids aspirated after OK-432 therapy [11, 14, 15]. When OK-432 is administered locally, inflammatory cells, such as neutrophils and monocytes, infiltrate the lesion and various cytokines are secreted [15]. These cytokines might induce very strong local inflammatory reactions in cystic disease, resulting in fluid drainage, shrinkage and fibrotic adhesion of cystic cavity [14, 15].

When cosmetic and economical aspects are considered, OK-432 therapy is superior to surgical procedures and can be implemented as an alternative to surgery [7, 10, 14]. Surgery should be considered only in a limited number of cases characterized by poor or no response to OK-432 treatment.

CONCLUSIONS

Our results suggest that OK-432 treatment of recurrent thyroglossal duct cyst after surgery is simple, easy, safe and effective. It can be used as a first-line treatment for recurrent cysts.

REFERENCES

- Anderson J.L., Vu K., Haidar Y.M., Kuan E.C., Tjoa T.: Risks and complications of thyroglossal duct cyst removal. *Laryngoscope*, 2020; 130(2): 381–384.
- Bratu I., Laberge J.M.: Day surgery for thyroglossal duct cyst excision: a safe alternative. *Pediatr Surg Int*, 2004; 20(9): 675–678.
- Qu R., Wang C., Dong Z., Li J., Liu D.: Another Strategy for the Treatment of Thyroglossal Duct Cyst: Totally Endoscopic Surgery by Breast Approach. *Surg Laparosc Endosc Percutan Tech*, 2018; 28(2): 118–122. doi: 10.1097/SLE.0000000000000514.
- El-Anwar M.W., Nofal A.A.: Thyroglossal duct cyst excision with hyoid bone preservation. *Eur Arch Otorhinolaryngol*, 2016; 273(6): 1521–1526. doi: 10.1007/s00405-015-3624-7.
- Lee D.W., Tae K.: Robot-assisted excision of thyroglossal duct cyst by a postauricular facelift approach. *Wideochir Inne Tech Maloinwazyjne*, 2020; 15(1): 245–248. doi: 10.5114/wiitm.2019.88751.
- Rohof D., Honings J., Theunisse H.J. et al.: Recurrences after thyroglossal duct cyst surgery: Results in 207 consecutive cases and review of the literature. *Head Neck*, 2015; 37(12): 1699–1704.
- Isaacson G., Kaplon A., Tint D.: Why Central neck dissection works (and fails) for recurrent thyroglossal duct remnants. *Ann Otol Rhinol Laryngol*, 2019; 128(11): 1041–1047.
- Cheng J., Lerebours R., Lee H.J.: Current trends and 30-day surgical outcomes for thyroglossal duct cyst excision in children. *Int J Pediatr Otorhinolaryngol*, 2020; 128: 109725.
- Ohta N., Fukase S., Watanabe T. et al.: Treatment of thyroglossal duct cysts by OK-432. *Laryngoscope*, 2012; 122: 131–133.
- Kusano Y, Ikeda R, Saito Y. et al.: Treatment of oral ranula in HIV-positive patient. *Auris Nasus Larynx*, 2021; 48(1): 171–174. doi: 10.1016/j.anl.2020.02.009.
- Ohta N., Fukase S., Suzuki Y., Kurakami K., Aoyagi M., Kakehata S.: OK-432 treatment of ranula extending to the parapharyngeal space. *Acta Otolaryngol*, 2014; 134(2): 206–210.
- Ohta N., Fukase S., Suzuki Y. et al.: Treatment of various otolaryngological cystic diseases by OK-432: its indications and limitations. *Laryngoscope*, 2010; 120: 2193–2196.
- Ohta N., Fukase S., Watanabe T. et al.: Effects and mechanism of OK-432 therapy in various neck cystic lesions. *Acta Otolaryngol*, 2010; 130: 1287–1292.
- Ohta N., Fukase S., Suzuki Y., Aoyagi M.: Treatment of salivary mucocele of the lower lip by OK-432. *Auris Nasus Larynx*, 2011; 38(2): 240–243.
- Ohta N., Fukase S., Kusano Y. et al.: Treatment of auricular hematomas by OK-432: How and why it works. *Otol Neurotol*, 2019; 40(8): e820–e823.
- Lekkerkerker I., van Heurn E.L., van der Steeg A.F., Derikx J.P.: Pediatric thyroglossal duct cyst: Post-operative complications. *Int J Pediatr Otorhinolaryngol*, 2019; 124: 14–17. doi: 10.1016/j.ijporl.2019.05.035.
- Batazzi A., Leng S., Ghionzoli M. et al.: Thyroglossal duct cyst: Factors affecting cosmetic outcome and recurrence. *Pediatr Int*, 2019; 61(10): 1020–1024. doi: 10.1111/ped.13955.
- Alahmadi A.A., Bawazir O.A., Rajab M.K. et al.: Thyroglossal duct surgery. What is the acceptable recurrence rate? *Saudi Med J*, 2020; 41(8): 878–882. doi: 10.15537/smj.2020.8.25169.
- Pastore V., Bartoli F.: “Extended” Sistrunk procedure in the treatment of recurrent thyroglossal duct cysts: a 10-year experience. *Int J Pediatr Otorhinolaryngol*, 2014; 78(9): 1534–1536.

Word count: 1315

Tables: 1

Figures: 1

References: 19

DOI: 10.5604/01.3001.0014.9073 Table of content: <https://otolaryngologypl.com/issue/14180>

Copyright: Some right reserved: Polish Society of Otorhinolaryngologists Head and Neck Surgeons. Published by Index Copernicus Sp. z o.o.

Competing interests: The authors declare that they have no competing interests.



The content of the journal „Polish Society of Otorhinolaryngologists Head and Neck Surgeons” is circulated on the basis of the Open Access which means free and limitless access to scientific data.



This material is available under the Creative Commons – Attribution-NonCommercial 4.0 International (CC BY-NC 4.0). The full terms of this license are available on: <https://creativecommons.org/licenses/by-nc/4.0/legalcode>

Corresponding author: Dr. Nobuo Ohta; Division of Otolaryngology, Tohoku Medical and Pharmaceutical University Hospital 1-12-1, Fukumuro, Miyaginoku, Sendai 983-8512, Japan; Phone: +81 22 259 1221; E-mail: noohta@hosp.tohoku-mpu.ac.jp

Cite this article as: Ohta N, Fukase S, Nakazumi M, Sato T, Suzuki T.: OK-432 treatment in pediatric patients with recurrent thyroglossal duct cyst after surgery; Otolaryngol Pol 2021; 75 (6): 28-32
