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Cohort Study Results of the one-stage proximal hypospadias repair with modified Koyanagi technique: A prospective cohort study in a single Vietnam centre

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ARTICLE INFO	A B S T R A C T			
A R T I C L E I N F O Keywords: Proximal hypospadias Koyanagi technique Modification HOSE score	Introduction: Proximal hypospadias is the most severe type of hypospadias. Our approach to Koyanagi technique for proximal hypospadias aims to improve the blood supply to the neourethral flaps and reduce meatal complications. <i>Methods:</i> Our prospective study included 75 patients who were operated for proximal hypospadias by our Koyanagi technique at Viet Duc hospital between January 2019 and December 2020. The clinical information obtained included a detailed medical history; preoperative, intraoperative, and postoperative data; short-term outcomes by the HOSE score were evaluated by a different physician. <i>Results:</i> The mean (range) age was 3.59 ± 2.41 years (1.5–14), 86.7% under 5 years old. There are 31 peno- scrotal, 31 scrotal, and 13 perineal hypospadias. The length of the neourethra ranged from 3.5 to 8 cm, mean 5.02 ± 0.88 cm. Evaluation of the surgeon at 6 months after surgery: primary success 81.3%. Complications occurred in 14 cases (18.7%), included 10 urethrocutaneous fistula and 4 dehiscence of the urethra. No cases of meatal stenosis or recession, urethral stricture, urethral diverticula. The mean HOSE score was found to be 14.47 ± 1.35 , ranged 11 to 16. 57 patients (76%) had a total HOSE 14 and above and 18 patients had score below 14 (24%) <i>Conclusion:</i> Our modified Koyanagi technique give us a good result for one-stage reconstruction of proximal hypospadias. Applying the HOSE score makes postoperative evaluation of hypospadias more objective and reliable.			

1. Introduction

Hypospadias is one of the most common congenital malformations of the male genitalia, occurs in approximately 1:300 newborn boys [1]. Proximal hypospadias is accounting for 20%–25% of all hypospadias, defined by a urethral meatus located at the penoscrotal junction, scrotum, and perineum after penile degloving in the operating room, is the most severe manifestation of the hypospadias spectrum [2]. These components include ventral penile curvature, penoscrotal transposition and bifid scrotum. Severe hypospadias is still a challenge for pediatric urologists, and there is still great disagreement on the ideal surgical approach with more than 300 procedures, especially about staged or one-stage operation [3]. One-stage options include the tubularized incised urethroplasty or various graft or flap-based techniques, which are an attractive option in that they may reduce cost, hospital stay, anesthetic risks, and time to the final result [2,3].

In 1983, Koyanagi et al. [4] developed a one-stage operation, for severe hypospadias, utilizing a perimeatal-based foreskin flap for the urethroplasty, but it was initially not widely used due to high complication rates (47,1%) [5]. For more than 30 years, many modifications of the Koyanagi technique have been introduced, with improved surgical outcome, complication rate about 20–25%, notably the authors' improvement such as Snow and Cartwrigh (1994) [6], Emir (2000) [7], Hayashi (2001) [8], Kang (2016) [9] ... These modified techniques in the literature were the purpose of improving the blood supply to the neourethral flaps and reduce postoperative complications. Each

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modification has its own advantages. We also tried to optimize the surgical techniques and apply them to our modified Koyanagi technique, to get better and better surgical results. The HOSE (Hypospadias Objective Scoring Evaluation) which was defined by Holland [10] in 2001, is a scoring system designed to allow an objective appraisal of the outcome of hypospadias repair. Many authors [11] have used the HOSE to make the postoperative evaluation of hypospadias more objective and reliable. This report aims to describe our approach to Koyanagi technique for proximal hypospadias at our hospital and short-term outcomes by the HOSE score. All our work has been reported in line with the STROCSS 2019 guidelines [12].

2. Materials and methods

Registration and ethics: Research Registry number is stated, in accordance with the declaration of Helsinki. Unique identifying number: researchregistry7272, available at https://www.researchregistry.com/browse-the-registry#home/registrationdetails/616c3a90 e2736c001fbbbff1/

2.1. Patients and methods

Our prospective cohort study recruited patients who underwent proximal hypospadias repair by our modified Koyanagi technique at Viet Duc Hospital (Hanoi) - one of the largest single centres in Vietnam from January 2019 to December 2020. The patient underwent surgery by the same surgical team.

Written informed consent was obtained from all patients before participation. The study was approved by our research committee, Viet Duc University Hospital, Hanoi, Vietnam and this was approved by Hanoi Medical University Institutional Ethical Review Board (No NCS07/BB-HĐĐĐ) date February 14, 2019.

Pre-operation: Previous medical and surgical history were collected. The karyotype was screened in order to rule out a primary chromosomal defect behind the disease of sexual differentiation. Pelvic ultrasound was performed for all patients to in the aim of searching for a prostatic utricle. Ages, glans, length of penis, location of the meatus preoperative, degree of chordee, penoscrotal transposition, and bifid scrotum were recorded.

Intra-operation: We collected parameters of operative time, location of the meatus after degloving, length of neourethra, cured the chordee by dorsal plication, phalloplasty.

Post-operative follow-up: All patients were followed by our pediatric team. Patients were followed up until July 2021. The patients were followed up with 3 visits at 1 month, 3 months, and 6 months. They were assessed for postoperative complications, a urinary stream, glans configuration, straightness of shaft, and completeness of transposition correction. The primary surgery is successful if the absence of any complications, uninterrupted urinary stream and good cosmetic after the first surgery. The patient was assessed by another physician by the HOSE [10] score at 6 postoperative months. The HOSE is a scoring system designed to allow an objective appraisal of the outcome of hypospadias repair, based on evaluating meatal location, meatal shape, urinary stream, straightness of erection, and urethral fistula.

2.2. Surgical techniques

A specialized surgical instrument for hypospadias, an 8–10 Fr silicone double-lumen balloon catheter was placed, 7/0 or $6/0 \times \text{long-term}$ absorbable monofilament sutures (PDSTm in our center) were used, the size of which depended on the development of the penis and the urethra, $3.5 \times \text{loupe}$ magnification. The procedure was performed under general anesthesia and caudal analgesia. The parameters of the length of the penis, the diameter of the glans was measured. A vertical stay suture was taken with polyprolene on the glans. A skin inner incision line which is drawn with a marking pen, is a circumferential incision was made 3–8 mm proximal to the coronary sulcus (dorsal penile glans from 3 mm sulcus, the incision on both sides is inclined downwards, forward, the ventral penile incision is the horizontal line from 5 to 8 mm coronary sulcus).

The foreskin and penile skin were degloved to the base of the penis between Buck's fascia and the dartos fascia. The urethral plate and the fibrous bands were separated to the base of the penile to correct the chordee and we dissected through the penoscrotal junction (Fig. 1A).

Performing an artificial erection test to check penile curvature (Fig. 1B). After resection of the fibrous bands on the ventral and penoscrotal penis, if the slightly and moderately chordee was still present, we were performed dorsal plication. Create a tunnel into the glans by dissecting the skin and glans separated from the corpus cavernosum, from the horizontal line to the top of the glans, to the sides, enough for the Hegar's dilator No. 6 of 7 to pass easily (depend on the size of glans) (Fig. 1C).

Incision in the middle of the foreskin skin (Fig. 1D), the thickness of the cut is slightly more than the anteroposterior diameter of the penis, move the 2 flaps towards the ventral penis (Fig. 1E). The inner edges of the flaps are sewn together by continuously stitching with 7/0 PDSTm. We have essentially created a neourethral plate that can be fixed to the penile albuginea by some stitches (Fig. 1F).

Drawing outer incision, each side often from 0.7 to 0.8 cm inner suture line. Our U-shaped outer incision is in front of the meatus, we divided it into short pieces, each about 2 cm. In each segment, we dissect the vascular peduncle to both sides just enough (Fig. 1G).

The neourethra was brought to the top of the glans through the glans tunnel. Urethral anastomosis with 7/0 PDSTm, some interrupted stitches. A pedicled flap (which was the tunica vaginalis or the scrotal dartos) was dissected carefully and the lateral edges of it were sewed to the ventral fascia of the penile by interrupted sutures. That way insured an additional coverage for the neourethra, which may reduce the postoperative complication rate (Fig. 1H).

We performed scrotoplasty and corrected the penoscrotal transposition and bifid scrotum if any. The Byar's flap created from the dorsal hood was used for the phalloplasty. Sometimes, the penis could be covered with scrotum skin if it was lacking (Fig. 1I). Note that the penile skin should not be stretched, or the flap was anemic.

A light compression dressing was used to cover the wound for some days. If there are no complications of bleeding, infiltration of fluid or urine into the incision, the dressing should be changed after 5 days. At our center, the patients were hospitalized, given antibiotics for 7 days, pain relief, care for the urinary catheter and remove on postoperative day 12–14.

Fig. 2, Fig. 3 and Fig. 4 shows some intraoperative photos.

3. Results

During the study period, a total of 75 boys with proximal hypospadias were operated on at Viet Duc hospital by the same modified Koyanagi technique. Patient age at the time of surgery ranged between 1.5 and 14 years, with a mean age of 3.59 \pm 2.41 years. 86.7% under 5 years old. In all cases, the meatus was at or proximal to the penoscrotal junction and was classified as 31 penoscrotal cases (41.3%), scrotal cases (41.3%), or 13 perineal cases (17.4%). Before penile degloving, all cases involved varying degrees of chordee, and 20 cases of penoscrotal transposition and 35 cases of the bifid scrotum with different degrees. Accompanying malformations: 7 patients with cardiovascular defects (1 Tetralogy of Fallot, 2 ventricular septal defects, 1 atrial septal defect, 1 mild tricuspid valve regurgitation, 1 Kawasaki syndrome, 1 pectus excavatum) was treated stably. 1 patient with esophageal atresia was operated on in the neonatal period, 1 patient with cleft palate underwent surgery, 2 patients with prostatic utricle underwent laparoscopic excision. The patients with unilateral undescended testicle (n = 11), bilateral undescended testicles (n = 2), unilateral inguinal hernia (n = 2)were surgically cured 3-6 months before the hypospadias repair and 1



Fig. 1. Our modified Koyanagi technique.

(A) Degloving to the base of the penis; (B) artificial erection and chordee correction; (C) Create a tunnel into the glans; (D) Incision at the midline of the dorsal prepuce; (E) Moving the 2 flaps towards the ventral penis; (F) Reconstruct neourethral plate; (G) Drawing outer incision, sufficient dissection and suture in small segments of anterior suture; (H) Protective intermediate layer by the tunica vaginalis or the scrotal dartos; (I) Phalloplasty and scrotoplasty.

patient with multiple malformations: anocutaneous fistula (underwent anorectal surgery at neonatal), right renal agenesis, right microtia ear, and prostatic utricle. 1 patient with chromosomal abnormalities (46XY 16qh (+)) and 5-alpha-reductase deficiency (n- = 5). Hormonotherapy was used in 10 patients preoperatively.

The operation time ranged from 120 to 180 min, mean of 145.9 \pm 17.5 min. The follow-up period ranged from 9 to 33 months. The length of the neourethra ranged from 3.5 to 8 cm, mean 5.02 \pm 0.88 cm. There were 15/75 cases (20%) that need to be cured the chordee by dorsal plication and 26/75 cases (34.7%) that need be covered with scrotal skin. Follow-up results at 6 months postoperatively: Primary success occurred in 61 cases (81.3%) with accepted cosmetic appearance and

adequate-sized glandular meatus. Classification of hypospadias in successful cases: 24 penoscrotal, 26 scrotal, and 11 perineal. The success rate of surgery was not related to the disease (p > 0.05). Complications occurred in 14 cases (18.7%). Urethrocutaneous fistula occurred in 10 cases (13.3%), dehiscence of the urethra occurred in 4 cases (5.4%). There was no incidence of meatal stenosis, meatal recession, urethral stricture, urethral diverticulum, rotation, or recurrent curvature. These complications were treated six months later.

The HOSE scores of the patients at 6 months after surgery were evaluated by a different physician (Table 1). The mean postoperative HOSE score was 14.47 ± 1.35 , ranged from 11 to 16 points. HOSE score classification: 57 patients (76%) in the good group with score 14 and



Fig. 2. Marking skin inner incision line, degloving and create a tunnel.



Fig. 3. Incision of the dorsal prepuce, neourethral plate and marking outer incision line.



Fig. 4. Anterior urethral suture lines and Phalloplasty.

Table 1	
Evaluation of surgical outcomes by the HOSE scores.	

Variable		Number of patients or mean
Meatal location	Distal glanular	45
	Proximal glanular	29
	Coronal	1
	Penile shaft	0
Meatal shape	Vertical slit	33
	Circular	42
Urinary stream	Single stream	63
	Spray	12
Erection	Straight	56
	Mild angulation ($<10^{\circ}$)	17
	Moderate angulation (>10° but	2
	<45°)	
	Severe angulation (>45°)	0
Fistula	None	61
	Single – subcoronal or more	10
	distal	
	Single - proximal	3
	Multiple or complex	1
Surgical results	Success $(n = 61)$	14.92 ± 0.94
	Failure ($n = 14$)	12.50 ± 1.09
Types of	Penoscrotal ($n = 31$)	14.39 ± 1.45
hypospadias	Scrotal ($n = 31$)	14.77 ± 1.17
	Perineal ($n = 13$)	13.92 ± 1.38

above, 18 patients (24%) in the average group with score 11 to 13, no patients were in the poor group (score below 11). Although no statistically significant difference was detected in HOSE score between the groups, the lowest HOSE score was found in the complication group.

4. Discussion

Hypospadias is one of the most common congenital malformations of the male genitalia [2]. Repair of proximal hypospadias involves correction of several components of the hypospadias complex with the intent of optimizing long-term functional and cosmetic outcomes. These components include ventral penile curvature greater than 30° , proximal location of the urethral meatus, ventral skin deficiency, abnormal glans morphology, abnormal division of the corpus spongiosum, penile torsion and penoscrotal transposition [10,11]. Proximal hypospadias is one of the most challenging conditions to correct. Although staged repair for severe hypospadias is a reliable option, several attempts have been made to do it in one stage [3], especially in a developing country like Vietnam. Because it's an attractive option for the patient, in that they may reduce cost, hospital stay, anesthetic risks, and time to the final result.

Koyanagi et al. [4] described a single-stage procedure whereby the urethroplasty with a parameatal-based and fully extended circumferential foreskin. Koyanagi et al. [5] reported a complication rate of 47% with good cosmetic results. The high complication rate is mainly due to poor blood supply especially the lateral side of the pedicle. However, many authors [7-9] have noted that the Koyanagi technique has some obvious advantages: there are no anastomoses between the neourethra and the original meatus, effectively reducing the rate of urethral stricture; the procedure always ensures that there is a sufficient flap for the neourethra; avoid the risk of penile torsion; the penoscrotal transposition or bifid scrotum can be corrected simultaneously. Snow and Cartwright (1994) [6] modified the Kovanagi repair, named "Yoke hypospadias repair", using an island-flap technique, which preserved a reliable dual blood supply to a long neourethra, but reported only 4 patients and complication rate was 50%. Koft et al. (2000) [7] described a modification of the technique which focuses on carefully preserving the lateral blood supply to the neourethral flaps, creates a sufficiently large urethral meatus equivalent to 20 Fr, their initial success rate of 80%. Hayashi et al. (2001) [8] also modified the Koyanagi repair by preserving a wide vascular pedicle for the distal skin flaps and by utilizing the distal urethral plate as the base for the distal neourethra. The overall success rate was 70% and a 15% rate (3/20 cases) of meatal stenosis. Sugita et al. (2001) [13] reported a series of 151 cases of severe hypospadias, their modified technique requires removal of the subcutaneous tissue of the distal portion of the flap, which acts as a full-thickness free skin graft and their total complication rate of only 15.9%, 3 cases (2%) meatal stenosis, 2 cases (1.3%) meatal drop-back. Hayashi (2007) [14] performed another modification that focused on reducing the incidence of meatal stenosis, they had 1/12 case (8%) with urethral fistula, no instance of meatal stenosis, or urethral stricture. Kang et al. (2016) [9] emphasized some following points in their modification that after curing the chordee, the urethral plate was recreated using foreskin, and then a U-shaped incision was made on the original and recreated urethral plate (as in the Duplay technique); a pedicled flap of the tunica vaginalis or scrotal dartos was used for additional coverage of the neourethra. Thanks to that their total complication rate was 20.8%. According to the (Table 2), recent publications reported the success rate of the modified Koyanagi repair

Table 2

Results of pro	oximal hypos	padias by F	Koyanagi te	chnique in	the literature
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Year published	Author	Procedure	Studied series	Overall success rate (%)	Complication
1994	Koyanagi et al. [5]	orginal	70	53	15 fistulas, 12 meatal stenosis, 6 stricture
1994	Snow and Cartwright	Yoke repair	4	50	2 fistulas
1996	Hayashi et al. [16]	orginal	17	52.8	3 Fistula, 3 meatal stenosis, 2 dehiscence
1998	Glassberg et al. [17]	orginal	14	50	7 fistulas
2000	Emir, Koff et al. [7]	Modified	20	80	4 fistulas
2001	Hayashi et al. [8]	Modified	20	70	3 fistulas, 3 meatal stenosis
2001	Sugital et al. [13]	Modified	151	84.1	19 Fistula, 3 meatal stenosis, 2 dehiscence
2003	Jorgensen et al. [18]	Modified	9	55.5	3 fistulas, 2 meatal stenosis
2007	Hayashi et al.	Modified	12	91.7	1 fistula
2009	Catti et al. [15]	orginal	26	38.5	11 Urethral dehiscence, 5 fistulas, 9 meatal or urethral stenosis, 7 urethrocele
		modified	31	38.7	6 Urethral dehiscence, 12 fistulas, 5 meatal or urethral stenosis, 5 urethrocele
2009	Elisangela de Mattos e Silva et al. [19]	Modified	26	38.5	Fistula, stenosis, urethral dehiscence, diverticulum
2010	Nerli et al. [20]	modified	14	64.3	3 Fistula, 1 meatal stenosis, 1 dehiscence
2010	Elsaied A. et al. [21]	Modified	30	90	2 fistulas, 1 meatal recession
2011	Arnaud et al. [22]	modified	21	23.8	Fistulas, urethral dehiscence
2013	Vepakomma et al. [23]	modified	24	54.2	Fistula, meatal stenosis, debiscence
2016	Kang et al. [9]	modified	24	79.2	4 Fistulas, 1 urethral
2017	Seleim et al. [24]	Modified Yoke	31	84	4 fistulas, 1 meatal drop-
2021	Akkary et al. [25]	Orginal modified	4 15	0 60	Fistula, stenosis, urethral dehiscence, diverticulum

varying from 60% to 92%. Some studies have shown that the Koyanagi technique was not as easy and had high complications. Catti et al. (2009) [15] reported 61.5% failure rate with the original Koyanagi technique and 61.3% failed with the Hayashi modification. In our study, the primary success occurred in 61 cases (81.3%) with accepted cosmetic appearance and adequate-sized glandular meatus. We had 18.7% complications rate which included 13.3% urethrocutaneous fistula and 5.4% dehiscence of the urethra. There was no incidence of meatal stenosis, meatal recession, urethral stricture, urethral diverticulum, rotation, or recurrent curvature. Compared with other authors (Table 2), the success rate in our study is high, although still lower than that of Sugital [14], Hayashi [17], Elsaied A [18].

Koyanagi technique [4] was characterized by 2 incisions (inner and outer) and 2 urethral suture lines (anterior and posterior). Some authors performed outer incision first, such as original Koyanagi technique [4], Yoke repair [6], modified Sugita [13]. They eliminated the penile curvature due to the skin before deciding whether urethral plate transection. However, most of the proximal hypospadias were accompanied severe chordee and some cases of a tethering urethral plate, this approach may also contribute to increased penile length [3]. As in the modified technique by Koff [7], Hayashi [8], Kang [9] ... in this study, we performed first the inner incision and penile degloving thereby eliminating the most cause of curvature. We used dorsal plication in 15/75 cases (20%). In our center, all cases of severe chordee which need a ventral penile lengthening, we will do 2 stages. We have followed the guidelines for the treatment of servere hypospadias [1,3].

At our hospital, we have employed the Hayashi modified Koyanagi technique for proximal hypospadias since 2012 and we found that there are difficulties or risk factors that easily lead to postoperative failure, which are: (1) The extensive dissection can easily injure the vascular pedicle, especially the lateral pedicle; (2) after flap surgery, the skin and mucosa of the foreskin tends to shrink, affecting the urethral suture even though we used $3.5 \times$ loupe magnification and the width of each side of the urethra was about 7–8 mm [8,9], leading to difficulties in creating the neourethra; (3) glans breakdown or complete breakdown was often occurred due to broken sutures, which were mainly caused by distension of the glans or the urinary catheter may be retracted to the ventral penis during postoperative care. That's why we have made small changes in our technique, which may just be the authors' experiential learning applications to combine for our modified technique to make the surgery easier. We emphasized the following points: (1) A full-thickness incision was made at the midline of the dorsal prepuce of the penis and The inner edges of the flaps are sutured early to reconstruct the urethral plate (same as stage 1 in multi-stage repairs), it was also emphasized by Kang's modification [9]; (2) Outer incision is made only through skin preserving underlying blood supply to the flap as in modified technique by Emir et al. [7] We divided outer incision into short pieces to keep the urethral plate from shrinking; (3) We didn't use the splitting the glans, we created a large tunnel enough for the Hegar's dilator No. 6 of 7 to pass easily (equivalent size Foley 18-22). This change reduced the meatal complication; (4) The additional coverage (the tunica vaginalis or scrotal dartos) on the neourethral which was reported in many studies [9,26] decreased the risk of urethrocutaneous fistula.

Aesthetic of our patients after hypospadias repair was evaluated using the HOSE score [10] by an independent urological surgeon. Many authors have published studies showing that the HOSE score is a reproducible objective outcome measure after hypospadias surgery, and independent of the initial severity of the defect. In our study, the mean postoperative HOSE score was 14.47 \pm 1.35, ranged from 11 to 16 points, the HOSE score of 14 and above in 76% of the patients. Since 86.7% of our patients were younger than 5 years of age, it was difficult to use uroflowmetry in that group, therefore, postoperative functional assessment by uroflowmetry was not used in this study. The modified Koyanagi technique has given us an excellent outcome for proximal hypospadias. In the future, we hope to have studies with larger numbers of patients. In 1995, John Duckett [2] introduced the term "hypospadiology", defined as the in-depth study of the art and science of the surgical correction of hypospadias, is currently flourishing. Thanks to the previous research of the hyposadiologists, it will help the next generation surgeons continue to research and get better results for this surgery.

The strength of our study is that there are a large number of patients with proximal hypospadias which underwent the same technique by the same surgical team within 2 years, contribute to increasing the research on the effectiveness of one-stage repair in general or Koyanagi technique in particular. However, the limitation of the study is that the follow-up time is short, and no functional test as uroflowmetry. We believed that a follow-up study with functional visits is need to evaluate long-term outcomes.

5. Conclusion

The Koyanagi technique remains a useful procedure for one-stage reconstruction of proximal hypospadias and gave good results. We have found modifications in our technique from the experience of many authors. Applying the HOSE score makes the postoperative evaluation of hypospadias more objective, reliable and help the next surgery better.

Ethical approval

Written informed consent was obtained from all patients before participation. The study was approved by our research committee, Viet Duc University Hospital, Hanoi, Vietnam and this was approved by Hanoi Medical University Institutional Ethical Review Board (No NCS07/BB-HĐĐĐ) date February 14, 2019.

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Author contribution

Tuan Hong Vu: operated the patients, designed the study, did the data collection, the data analysis, wrote manuscript.

Hoa Viet Nguyen: operated the patients, designed the study, revised the manuscript.

Quan Quy Hong: operated the patients, wrote manuscript.

Hung Quang Pham, Tung Thanh Pham: did the data collection. Dang Hai Do: Revised the manuscript.

Thanh Truong Do: Designed the study, revised the manuscript. All authors have read and approved the final version of the manuscript.

Registration of research studies

- 1. Name of the registry: Research Registry
- 2. Unique Identifying number or registration ID: researchregistry7272
- 3. Hyperlink to your specific registration (must be publicly accessible and will be checked): https://www.researchregistry.com/registe r-now#user-researchregistry/registerresearchdetails/616c3a90e 2736c001fbbbff1/

Guarantor

Dr. Tuan Hong Vu.

Consent

The publication of this study has been consented by the relevant patient. Informed consent was obtained from the parents for the report publication.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

None.

Acknowledgments

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.103012.

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