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Table of Contents

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Vectra 3D Imaging for Quantitative Volumetric Analysis of the Efficacy of Propranolol in Infantile Hemangioma

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Abstract

Infantile hemangioma (IH) is a common tumor in infants. After the proliferative phase, hemangiomas regress, but sometimes leave scars. Propranolol is now the recommended first-line oral therapy for IH. To evaluate the effectiveness of oral propranolol administration, we measured quantitative changes in an IH by digital camera and Computed Tomography (CT) or Magnetic Resonance Imaging (MRI). Although digital cameras are very simple, changes in color tones occur with time, lesions are evaluated in two dimensions, and changes in the thickness of the hemangioma are difficult to evaluate. Therefore, we investigated the feasibility of 3D photography to quantify volumes in IH during oral propranolol treatment.

Keywords

Infantile Hemangioma, Propranolol, 3D Photography

1. Introduction

Infantile hemangiomas (IHs) rapidly increase in size from birth until 5 months of age [1]. After the proliferative phase, the color of most IHs fades and the size decreases. Because it takes a long time for the color of IHs to fade and residual lesions have a wrinkle-like appearance, it is sometimes necessary to surgically remove scars. The effectiveness of oral propranolol in the treatment of IHs was first reported in Europe in 2008 [2], and a global randomized control study was

conducted [3] [4]. Propranolol is now the recommended first-line oral therapy for IH. The oral administration of propranolol for treatment of IH has been covered by national insurance in Japan since 2017. Before national insurance approval, we offered this treatment in our hospital after review and approved by the Ethical Review Board of Keio University School of Medicine (approval code No. 20130027).

To evaluate the effectiveness of propranolol oral administration, we first measured quantitative changes in IHs by digital camera and CT or MRI. Although digital cameras are very simple, changes in color tones occur over time, lesions are evaluated in two dimensions, and changes in the thickness of IHs are difficult to evaluate. Because the indication was limited by expense and radiation exposure, there were many patients with superficial lesions who were ineligible, but there were several patients who were eligible, such as those with an IH in the orbital space or huge and deep IHs. To overcome the shortcomings, we used a 3D camera (Vectra® H1; Canfield Scientific, Inc., NJ, USA) to measure quantitative changes in IHs. In this study, we determined whether or not 3D photography is a useful tool for measuring the surface area and volume of IHs over time during treatment.

2. Patients and Methods

This study included 10 patients treated with oral propranolol treatment from 2015-2016 in our hospital before propranolol treatment was covered by insurance in Japan. All IHs were in the proliferative phase and 3 months after. Oral propranolol treatment was provided for those who were expected to cause serious dysfunction such as respiratory and visual function or expected to have surgical resection after spontaneous regression, and for IHs in areas likely to be wedge-shaped (nose, lips, auricle, intercostal, etc.). The patients signed informed consult. Baseline screening by pediatric cardiologists included blood pressure, heart rate, hematologic examination, an echocardiogram, and an electrocardiogram. We initiated propranolol treatment in the hospital at a dose of 1 mg/kg per day and increased the dose to 2 mg/kg per day. After discharge from the hospital, we followed the patients in the outpatient clinic. We slowly tapered the 2 mg/kg per day dose after 6 months and stop the medication. 3D photographs of the IHs were taken using a 3D camera (Vectra® H1) before starting the medication, and 1, 3, and 6 months after starting the medication). The Vectra H1 is a handy 3D camera system; the images are easy to analyze and the volume measurements are easy to obtain. Because the 3D camera system is designed to take pictures of people, the shooting time is 0.002 seconds, the color image is highly accurate, and background noise is suppressed. 3D reconstruction was generated using the attachment software for the 3D camera. The IH lesion outlines were manually traced to automatically calculate the surface area and volume.

3. Ethical Considerations

This study was reviewed and approved by the Ethical Review Board of Keio

University before patient enrollment and data collection (approval code No. 20130027). The purpose of the study was explained in detail and the participants' parents voluntarily submitted a signed, written informed consent. Careful consideration was paid to protecting the privacy and personal information of the subjects.

4. Results

The average age at starting medication was 3.5 months. The study cohort consisted of 2 boys and 8 girls (Table 1). We calculated the IH surface area and volume at the start of treatment, and 1, 3, and 6 months after starting medication and followed up until three years old (1 case was lost to follow-up 1 month after starting medication and 2 cases were lost to follow-up 3 months after starting medication). The complication of treatments (hypotension, bradycardia, hypoglycemia, hyperkalemia, wheezing) does not occur. Case 1 was a 5-month-old boy. Two weeks after birth he developed an IH on the right chest and was referred to our hospital for oral treatment. Oral medication with a β--blocker was initiated at 2 mg/kg/day according to our protocol. 3D photographs of the IH were obtained before starting medication, and 1, 3, and 6 months after starting medication using a 3D camera (Vectra® H1). A 3D image before starting medication is shown in Figure 1. By 6 months after starting medication, the color of the IH had faded, but it was difficult to determine the surface area and volume of the IH using a 2D digital camera (Figure 2(a) and Figure 2(b)). We simulated the chest wall using 3D photography from the 3D coordinates of the IH margin and calculated the volume of the IH (Figure 3(a)). The volume was 23.3 cc. Six months after starting oral administration, the volume of the IH was 5.4 cc (Figure 3(b)). We also calculated the change in IH over time using the following formula: IH volume 1, 3, or 6 months after treatment (cc)/original IH volume (cc) × 100 (%). The volume of the IH lesion was significantly decreased during treatment (40.86% ± 17.19% at 1 month, 25.06% ± 14.41% at 3 months, and 8.69% ± 7.29% at 6 months; $p < 0.001$; Figure 4). The surface area of the IH was significantly decreased during treatment (82.23% ± 13.68% at 1 month, $p < 0.01$; 78.09% ± 28.79% at 3 months, $p < 0.05$; and 64.72% ± 22.36% at 6 months, $p < 0.001$; Figure 4). Case 2 is a 3-month-old girl. The change in IH over time is shown in Figure 5.

5. Discussion

A β-blocker has fewer side effects in treating IHs compared to steroids and interferon; especially good results have been reported in IHs involving the head and neck [5]. Although the mechanism of action of propranolol in treating IHs is not clear, propranolol is a non-selective beta-adrenergic antagonist with an affinity for β-1 and β-2 receptors. Propanolol causes down-regulation of bFGF and VEGF and induces apoptosis of capillary endothelial cells [6]. It has also been reported that propanolol selectively inhibits MMP-9 [6]. Propranolol has effica-

cy in the treatment of proliferating IH and facilitating the involution phase early, thus possibly avoiding surgery.

Using a 3D camera (Vectra® H1) is as easy as using a digital camera. Indeed, a 3D camera can easily measure the volume of IH lesions. 3D measurement is non-invasive, does not require radiation exposure and sedation as do CT and MRI, and is very useful in evaluating the treatment effect in infants. The reliability of 3D photography for IH volume changes over time (3 months after taking medication) has been reported [7]. Our study showed the volume and surface area changes in IHs by 6 months, as determined by 3D imaging. We should also consider the surface area. In case 3, a 4-month-old girl, even though no change in surface area was demonstrated, a decrease in volume was observed (**Figure 6**). It is our opinion that quantitative evaluation of IHs using a 3D camera is useful.

Table 1. Data of 2 boys and 8 girls.

No.	Location	Age	Sex	Weight	Recurrence	Adverse Event
1	Chest	6 m	M	7.2	-	-
2	Jaw	3 m	F	5.4	-	-
3	Cheek	3 m	F	6.9	-	-
4	Lip	3 m	M	5.9	-	-
5	Eyelid	3 m	F	6.1	-	-
6	Eyelid	4 m	F	5.3	-	-
7	Eyelid	3 m	F	5.5	-	-
8	Nose	3 m	F	5.7	-	-
9	Scalp	3 m	F	6.6	-	-
10	Eyelid	4 m	F	6.4	-	-

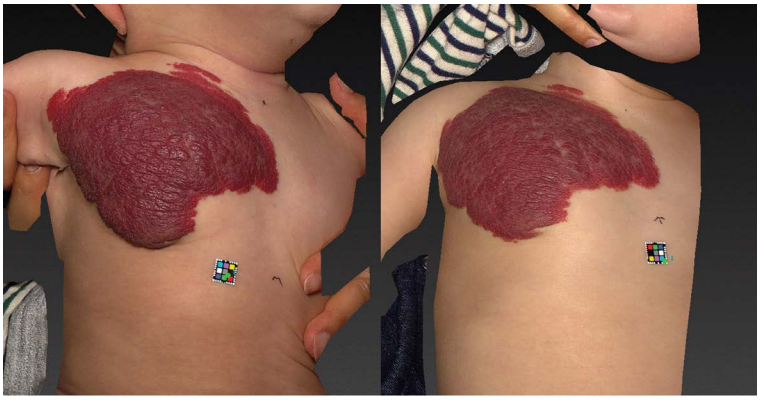
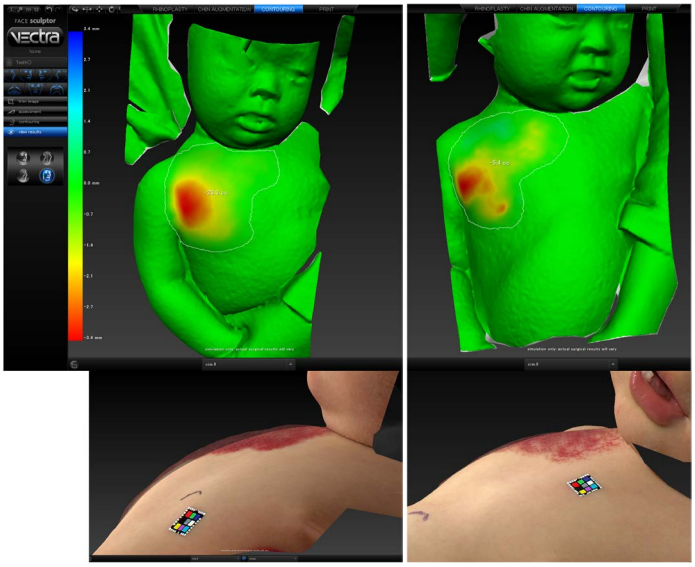


Figure 1. 3D imaging of IH in a 5-month-old boy (movie). The chest wall is simulated from the 3D coordinates of the hemangioma margin, and the volume of the hemangioma present on it is calculated.

(a) (b)

Figure 2. Photographs by 2D camera of a 5-month-old boy. (a) before starting medication; (b) after 6 months of propranolol treatment. Although these images show the change of flattens and fades, it is difficult to quantify the change in volume.



(a) (b)

Figure 3. Volumetric measurement using 3D photography of an IH in a 5-month-old boy. (a) before starting medication; Volume: 23.3 cc, Height difference between chest wall and hemangioma: 6 - 10 mm. (b) after 6 months of propranolol treatment. Volume: 5.4 cc, Height difference between chest wall and hemangioma: 2 - 3.5 mm.

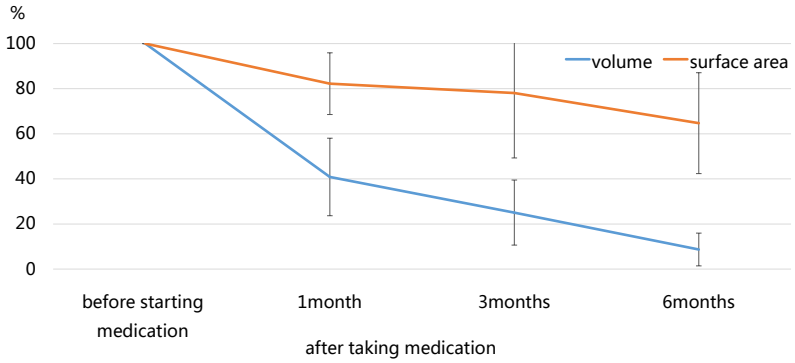


Figure 4. Volume and surface area change of IH during treatment. It shows the usefulness of the evaluation with a 3D camera.

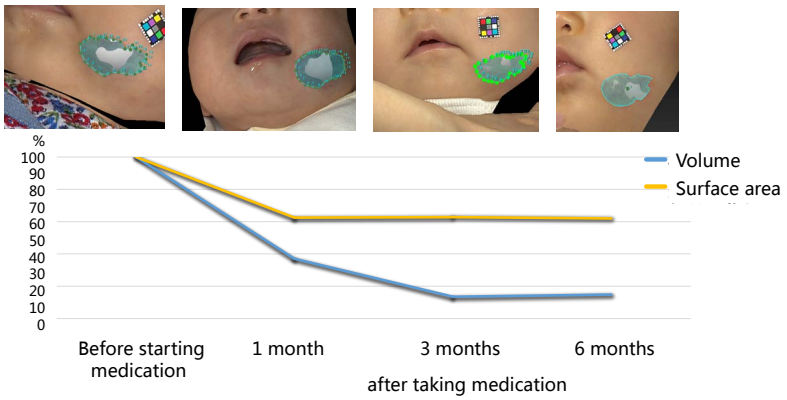


Figure 5. Changes in volume and surface area of IH during treatment in a 3 month-old girl. Although the surface area has not decreased, the volume has decreased by 80%. It shows the usefulness of the evaluation with a 3D camera.

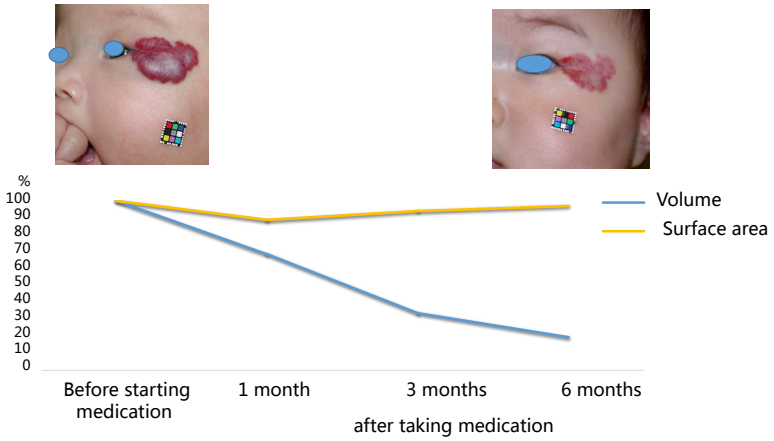


Figure 6. Changes in volume and surface area of IH during treatment in a 4-month-old girl. It shows that the surface area does not change much, but the volume is clearly reduced and improved with a 3D camera.

In this study, we followed infants for 6 months by using a 3D camera to measure the surface area and volume of IHs. The IH volume was shown to be greatly reduced with propranolol treatment compared with baseline. This result showed the usefulness of evaluation with a 3D camera. Even in cases in which the surface area did not change, the volume clearly decreased. A drawback of 3D imaging is that the shape of the lesion and simulation of the normal form may change and the camera is expensive. Nevertheless, we suggest that volumetric measurement of IHs using a 3D camera may be useful to evaluate treatment.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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Patients Body Modeling: A Practical Theoretical Experience in Plastic Surgery

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Abstract

This research is an active pedagogical experience with surgeons who are specialized in reconstructive plastic surgery and nursing. The guiding questions were the statements of each Surgeon's clients and what they think about them. Modeling the imaginary bodies of the patients and identify what they built in their minds, highlighting topics and the discussion about the practice made in clay modeling. Eight bodies were modeled, with six bodies with all the senses and two incomplete bodies with only the regions of the breasts and buttocks. It was concluded that there are diverse issues emerging such as gender, ethics, care, spirituality and dream as the fulfillment of the wishes of the patients. Where in the body can these surgeons act to improve the patients' physical and quality of life? There is an important theme when we are generally welcoming women into an anamnesis that considers their body as a whole, but what is their expectation for each patient? This paper showed the importance of the preoperative evaluation of this whole, for the indication of repairs or contraindications of procedures that aim to improve the physical of the patients, with possible bodily surgical modifications as a form of modeling through liposuction, grafting or excision with lipectomy and withdrawal of excess skin to mitigate the effects of the transformations that deform the physical and end up harming the human relationship, especially women. This is a qualitative method showing the meanings of the bodies of the women represented in the modeling by the Surgeons. However, some surgeons stand out by the look of their practice, as greater sensitivity looking at the body as a whole, spiritual and emotional. They need to use their skills as surgical art and gift, to try to achieve a means, but not an end.

Keywords

Modeling, Bodies, Surgery, Plastic, Art

1. Introduction

We have been working all our professional life operating patients who need surgical repairs in hospitals, with medical faculty and students interested in learning the plastic surgery area. As the world changes, everything around us also changes, as a way of understanding, teaching, and changing the way we do during the post-graduation course of plastic surgery by the Universities. We decided to use an atypical pedagogy, modeling the bodies of patients discussing life, not just the contents that involve teaching. We think that plastic surgeons develop practical and mental skills as they are able to model breasts, abdomen, nose, buttocks, etc. We often model subjectively without realizing how this happens. Often, young plastic surgeons apply a rational practice that marks or removes scars such as tattoos that are difficult to remove, remembering that these surgeons are not always able to achieve the desired result of patients' fantasies or delusions, since most of the time, we only reshape soft bone structures. In cases of birth defects or diseases as the main focus, there is sometimes a body modeling exaggeration for the permanence of eternal youth. As Gabriel Marcel affirms (page 3), there are no limits to the reality of the world. In this sense, the research question focuses on healthy bodies that want to be more perfect as an extension of youth and remaining healthy. At the moment, there is a privileged appreciation of self-centered "beautiful bodies", thinking that plastic surgery can accomplish dreams and bodily miracles. In this paper, the bodies were modeled in clay in the illusion of perfection. We actually try physical practices without considering the psychological, psychosocial, psycho-environmental, and psycho-spiritual dimensions. When thinking, we can fulfill our patients' dreams and desires for a miracle [1]. In Brazil, we currently have around 6083 Plastic Surgeons, considering that modeling images give meaning to what is represented from what surgeons think, but may or may not achieve.

This paper is related to the plastic surgeon's reaction to his patient's body and his attempt to model it in a clay sculpture to reflect, also, his patient's opinion of her own body. The objective is to choose the best surgical technique to meet the patient's expectations, though it is not always possible.

2. Material and Method

The qualitative study tries (according to Minaio, 2002) to work with a meaning attributed by the Surgeons to the facts, practical relationships and social phenomena, to interpret the practices and how to model the bodies through surgeries [2]. Portions of clay were distributed in the same amount so each doctor would

model the bodies of patients of their minds according to their conviction. The modeling was done in the classroom, which we consider as a living laboratory for the development and production of knowledge. The participants were post-graduate doctors with restorative training in Universities and Hospitals. Each doctor had an hour to turn a piece of clay into a body figure of a woman who came into their minds. The results are in **Figures 1-7**, without identification of the authors, but with the summary of what they thought [3].

Imaginary observations of 8 patients:

Figure 1. "I saw my patient in the mirror saying that she was horrible, fat and that she wants to keep her body in the form of a guitar, without stretching marks, without marks, and thin."

Figure 2. "I heard my patient saying: I want my breasts hard and round, without belly and attractive for my husband."

Figure 3. "My patient said, Doctor, it's no use having a pretty face and my body is deformed."

Figure 4. "I heard: 'As my doctor, I would like you to perform a miracle for me to be perfect.'"

Figure 5. "Doctor: I am here for you to model me, I want the woman's wishes fulfilled so that I have a new quality of life."

Figure 6. "I thought you could transform me. I keep my feet on the ground, but I consider you a God. I look at my body (A) in the mirror and I wish I had pointed breasts and a well-defined abdomen (B)."

Figure 7. "I would like to be transformed into perfection, but I do not know if you are an artist, I only know that you are a Plastic Surgeon."

Observation without a figure: "I saw on the Internet that you achieve the impossible, so I saved all my money, but I want to demand perfection."

Random phrases more heard in doctor's offices:

1) I hate to look at me in the mirror.
2) I feel horrible.
3) I want to have the face of the Italian artist.
4) After I had my children I was deformed.
5) I am saving money to become a guitar.
6) I want to be without marks and thin.

7) My breasts are crooked, but I want them round without a scar.
8) I want to put my whole body in the right place.
9) I want you to give me what I cannot get with other doctors.
10) I bring this photo, but I want to be like it.

The Organization suggests the dream: The beauty of the body, the wishes of the mind and soul brings the transformational possibility realized by the Surgeon (J. H. Resende, 2016) [4].

3. Results

We could divide the results of the study into two parts:

1) The surgeon physician evaluates the client's body such as:

- OK, no corrections needed.
- The breasts are in a pendulum and need to be lifted.
- The breasts are sagged but only with the prostheses corrected.
- The breasts do not need correction but liposuction would be necessary.
- Do the necessary corrections match the wishes of the customers?
- Should I operate this client?

2) Customers evaluate their own bodies, such as:

- I'm horrible!
- I need an urgent plastic surgery.
- How my tits are down!
- Do I want my 45-year-old body just like I was in 18?!
- I will break this mirror or make a plastic.
- I want to be beautiful and wonderful!

4. Analysis and Discussion

Through this kind of artistic approach (clay modeled bodies) the plastic surgeon exposes his practical reflections on the soul of these bodies. These women, as our patients, whish and dream of a miracle therapy. All this involves sensitivity, subjectivity, talent and good practice with much anticipation [5]. Velloso (2009) said: "It is a social and subjective construction of the body that challenges researchers from different areas of knowledge and also the artists who from the most different languages punctuate the text, engraving, sculpture or cinema that they try to express, interpret it and interpret it on human conditions …" [6], remembering that not always the dream will be able to come true! Also, the dangers inherent in a surgical act, even with a safe and careful preoperative.

5. Conclusions

Within the limit of pages to be published, this study has given us explanations of our knowledge about the body besides plastic surgery. We considered our care, our responsibilities, ethical and aesthetic [7]. It is necessary for these women who wish to undergo plastic surgery to keep "their feet on the ground". Goette said: "Healing without miracles and doing miracles silently."

The bodies were modeled in clay in this study to bring the illusion of perfection. What is perfection for a deformed body? [8] We conclude that plastic surgery sometimes can't transform a client in their expectations. We advise surgeons to do a pre-operative with explanations about what the client wants and what he can offer. This study is a reflection on the interdependence of factors such as technical knowledge of the plastic surgeon, his natural talent, and the state of each patient, whose body and mental health may cause possible intercurrences, which may affect the final results.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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Four Cousins with Hypospadias, Is It Familial or Environmental? (Case Report)

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Abstract

Hypospadias is one of the most common birth defects. However, its etiology remains largely unknown. Genetic and also intrauterine environmental factors have a principal role in causing familial hypospadias. Hypospadias repair is done through several techniques, one of which is Snodgrass technique which has many advantages. The 4 cousins underwent repair with no complications during 6 months follow up period. **Methods:** Urethroplasty using Snodgrass hypospadias repair of 4 cousins with positive parental consanguinity and absent family history of hypospadias. **Results:** 4 cousins underwent Snodgrass (Tubularized incised plate urethroplasty) with no complications after 6 months follow up. **Conclusion:** Although there is high heritability of hypospadias that also aggregates within second- and third-degree relatives, environmental factors may play a principal role in causing familial hypospadias. Hypospadias repair has a long learning curve. The continuous auditing should be done to improve the results.

Keywords

Hypospadias, Familial, Environmental, Snodgrass Repair

1. Introduction

Hypospadias is the most common congenital abnormality affecting the male urethra, with a worldwide incidence of about 1:300 live male births. Hypospadias is the abnormal location of the urethra on the ventral surface of the penis [1] [2].

The etiology of hypospadias in the majority of cases remains unknown. There is a genetic background for hypospadias through studying and observing genetic syndromes associated with hypospadias, genetic defects in the biosynthesis and function of Androgen and the mood of monogenic inheritance in families with

hypospadias [3] [4] [5] [6].

However, fewer than 5 percent of all cases of hypospadias may be associated with such conditions. In addition to a family history of hypospadias, only a few risk factors associated with hypospadias have been found, for example, paternal subfertility and intrauterine growth retardation [7] [8] [9].

The goal of hypospadias repair is to create a straight penis with a slit-like meatus at the tip of the glans and a urethra of uniform caliber and adequate length, reconstructing a symmetrical glans and penile shaft and achieving projectile stream and normal erection [10].

2. Case Report

Four cousin children were presented to our outpatient clinic, complaining of dribbling of urine and inability to pass a forward urine stream.

From history, there is positive consanguinity as their parents were 2 sisters (with a history of parental consanguinity) married to 2 brothers. No family history of similar conditions, nor medication history during pregnancy (See **Table 1**).

The first parents have 5 sons, 2 of them were with hypospadias; the first child was 10 years old, with distal penile hypospadias, his brother was 2.5 years old with sub-coronal hypospadias.

The second parents have 7 children, 3 of them are boys, 2 with hypospadias; the first boy was 9 years old with sub-coronal hypospadias. His brother was 2.5 years old with glanular hypospadias.

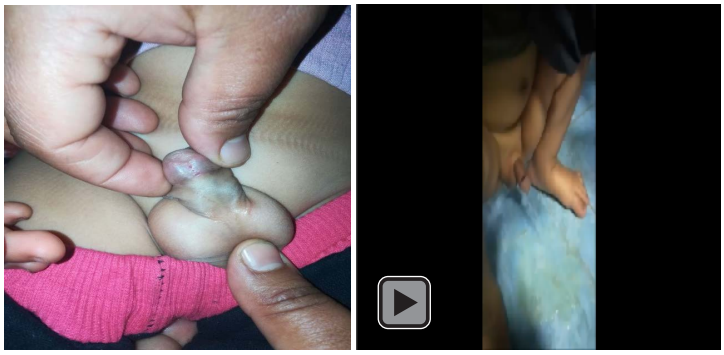
The 4 children underwent Snodgrass repair, in 3 of them we used preputial flap and the 4th one Dartos flap as a second intervening layer.

Post-operative hospital stay was 10 days, Catheter removed before discharge, urethral dilatation started immediately with frequency of once daily for the first week, then once weekly for the $_{\text{next}}$ month. Follow up was done once weekly. There were no complications for 6 months follow up (See **Cases 1-4**).

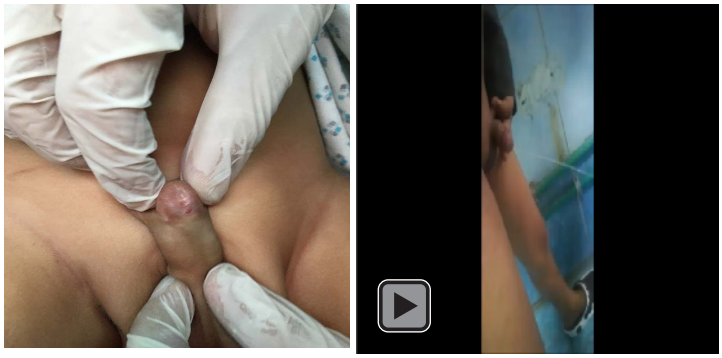
Table 1. Showing patient data and procedures done.

	Patient (1)	Patient (2)	Patient (3)	Patient (4)
Age	10 years	2.5 years	9 years	2.5 years
Relationship	Brothers		Brothers	
Diagnosis	Distal penile	Sub-coronal	Sub-coronal	Glanular
Chordee	Absent	Absent	Absent	Absent
Prepuce	Incomplete	Incomplete	Incomplete	Incomplete
Associated anomalies	Not present	Not present	Not present	Not present
Shape of the scrotum	Normal	Normal	Normal	Normal
Flap (Second layer)	Preputial	Preputial	Preputial	Dartos
Complications	UTI (treated)	-------	------	------

Case 1. Distal penile pre- and post-operative photos.



Case 2. Sub-coronal hypospadias pre-operative photo and post-operative video.



Case 3. Sub-coronal hypospadias pre- and post-operative photos and video.

Case 4. Glanular hypospadias.

3. Discussion

The inheritance appears to be transmitted equally through the paternal and maternal sides of a family. In studies of twin pairs, Kallen *et al.* [11] found a concordance rate of 18 percent, whereas another study reported concordance rates of 9 percent and 27 percent for dizygotic and monozygotic twins, respectively [12].

Brothers of a hypospadias case have, in smaller descriptive or case-control studies, been estimated to have about a 9 percent chance of developing hypospadias. It was found that the more severe the hypospadias in an individual, the higher the incidence in first-degree relatives, ranging from 3.5 percent to 16.7 percent [13].

It was found that hypospadias in second- and third-degree relatives was inherited equally from the maternal and the paternal sides. It has previously been hypothesized that hypospadias is one symptom of the testicular dysgenesis syndrome, which may be increasingly common because of adverse environmental influences, for example, estrogenic or anti-androgenic substances [14].

Here, consanguinity might be a cause of familial hypospadias and it was apparent in the second-generation members without previous history of similar conditions, so parents were notified with the increased risk of having grandsons with hypospadias and good prenatal care was advised in the future.

4. Conclusion

Although there is high heritability of hypospadias that also aggregates within the second and third-degree relatives, environmental factors may play a principal role in causing familial hypospadias.

Hypospadias repair has a long learning curve. The continuous auditing should be done to improve the results.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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