Case Report

Evaluation of the scrotal thermal environment and the testicular sizes in prepubertal hemiscrotal agenesis without cryptorchidism

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Abbreviations & Acronyms
HSA = hemiscrotal agenesis
PELVIS = perineal hemangioma, external genitalia malformations, lipomyelomeningocele, vesicorenal abnormalities, imperforate anus, and skin tag

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Introduction: Hemiscrotal agenesis is a rare scrotal developmental disorder. Orchiopexy on the opposite side of the scrotum with rugae or scrotoplasty combined with orchiopexy is usually performed as a surgical treatment for hemiscrotal agenesis with cryptorchidism. Till date, there are only eight published case studies of hemiscrotal agenesis.

Case presentation: A 6-year-old boy, who had been previously treated for infantile hemangiomas, was referred by a pediatrician to our hospital for the follow-up of hemiscrotal agenesis without cryptorchidism. Thermography demonstrated that the temperature of the right scrotal skin with no rugae was higher than that of the left scrotal skin with rugae. The patient's parent declined scrotoplasty because the hemiscrotal agenesis was to be followed up without scrotoplasty and scrotal ultrasonography revealed no abnormal findings in both testes.

Conclusion: High-temperature environment may not impair the testicular growth in prepubertal hemiscrotal agenesis without cryptorchidism.

Key words: hemiscrotal agenesis, PELVIS syndrome, thermal environment, thermography, ultrasonography.

Keynote message

High-temperature environment is thought to impair the testicular growth. In the present case of hemiscrotal agenesis, scrotal ultrasonography revealed no abnormal findings in right testes though the right scrotum was in high-temperature environment; thus, the patient's parent declined scrotoplasty. This is the first report of hemiscrotal agenesis without cryptorchidism followed up without surgical treatment until school age.

Introduction

HSA is one of the rarest anomalies among scrotal developmental disorders and is characterized by the unilateral absence of scrotal rugae in the perineum, between the penis and anus.[1] In complicated cases of cryptorchidism, scrotoplasty is often performed together with orchiopexy as a surgical treatment to improve scrotal appearance or the thermal environment of the testis.[2] However, surgical indications for HSA have not been clearly defined, and the decision to perform scrotoplasty is based on the preferences of the child's family.

Herein, we report a case of a 6-year-old boy with HSA without cryptorchidism who was followed up without surgical treatment until school age.

Case presentation

A 6-year-old boy was referred by a pediatrician to our hospital for follow-up of HSA. At birth, the scrotal rugae and pigmentation in the right scrotal skin were absent, and an infantile hemangioma was found in the right scrotal skin (Fig. 1a). Additionally, multiple infantile

hemangiomas were found, spread diffusely over the right lower limb (Fig. 1b). No obvious abnormal findings were observed on the spinal cord and pelvic magnetic resonance imaging. The patient was diagnosed with PELVIS syndrome, which was attributed to perineal hemangioma and malformations of external genitalia.[3] The infantile hemangioma of the right scrotum disappeared spontaneously, and the infantile hemangiomas of the right lower limb were treated using laser therapy.

Physical examination revealed that the right scrotum was flat and right scrotal rugae and pigmentation were absent (Fig. 2a). A midline raphe was present, and penile morphology was normal. The left testis was localized in the normal left scrotum, and the right testis was in the flat right scrotum lined with the non-rugae scrotal skin. Abdominal ultrasonography revealed no abnormal findings in the urinary tract.

A thermography camera (FLIR C5®; Teledyne FLIR LLC, Wilsonville, OR, USA) was used to assess the thermal environment around the scrotum. Thermography showed that the temperature of the flat right scrotum with right non-rugae skin and the left rounded scrotum with rugae skin were 34.9 and 33.9°C, respectively (Fig. 2b). Moreover, the temperature of the femoral skin adjacent to the right scrotum was high, measuring 36.4°C, while the temperature of the foreskin was low.

Assessment of the testicular growth using scrotal ultrasonography showed that both right and left testes were normal in size and echotexture (Fig. 3a,b). Moreover, color Doppler ultrasonography showed normal blood flow in the testes (Fig. 3c,d).

The patient's parents had been previously advised by the pediatrician that the patient's HSA would be followed up clinically, and no scrotoplasty would be performed; furthermore, scrotal ultrasonography revealed no abnormal findings in both testes. Therefore, the patient's parent declined scrotoplasty.

Discussion

HSA is the rarest anomaly among scrotal developmental disorders,[1] and only eight cases of HSA have been reported.[1,2,4–9] In two of these cases with normal descended testes, scrotoplasty was not performed.[1,4] Conversely, among the other six cases of HSA with cryptorchidism, four cases underwent scrotoplasty combined with orchiopexy,[2,5,7,8] and two cases underwent orchiopexy on the opposite side of the scrotum with rugae without scrotoplasty.[6,9] Thus, all six testes (four undescended and two ectopic testes) were covered with rugae scrotal skin, with or without scrotoplasty. Although there is no consensus on the indications for scrotoplasty, these surgeries were probably performed to provide an optimal thermal environment for the testes and prevent future impairment of testicular function.

The temperature of the undescended testis was significantly higher (34.4 ± 0.9°C) than that of the contralateral normally descended testis (33.2 ± 1.2°C; $p < 0.001$).[10] Moreover, because of the linear relationship between the position of testes and temperature, this temperature rise could either be a concomitant or the main factor causing testicular function impairment associated with cryptorchidism.[10] A previous study reported that the higher the position of the undescended testis, the smaller the size of the testis.[11] In addition, it has been reported that a decrease in testicular temperature is associated with postoperative testicular development.[12]

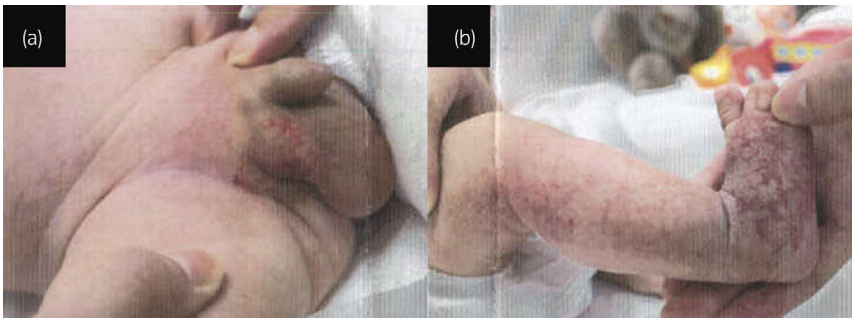

Fig. 1 Postnatal findings of hemangiomas. (a) Hemangiomas on the right scrotal skin with no rugae. (b) Diffuse hemangiomas from the right lower limb to the back of the right foot.

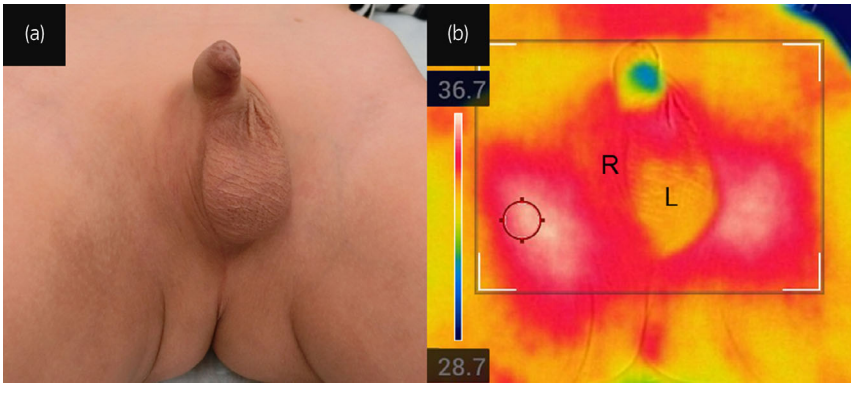

Fig. 2 External genitalia and thermography findings. (a) A midline raphe is present. There are no rugae on the right scrotal skin, although rugae are present on the left scrotal skin. Both right and left testes have descended. (b) Thermography showed that the temperature of the right scrotum (R) and the left scrotum (L) were 34.9 and 33.9°C, respectively. The temperature of the femoral skin, which is just outside of the right scrotum (red circle), was high, specifically 36.4°C, while the temperature of the foreskin was low.

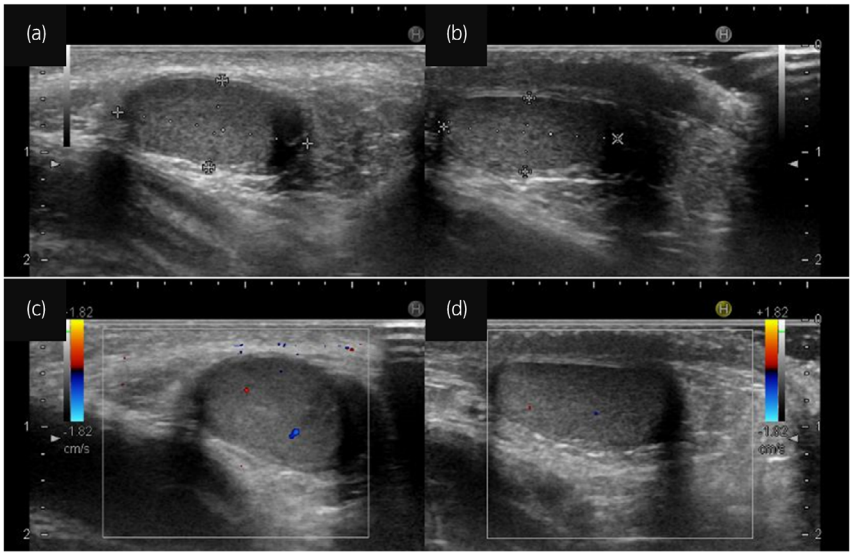

Fig. 3 Findings of scrotal ultrasonography. (a, b) Ultrasonography reveals the sizes of the right and left testes to be 18 × 8.7 mm and 16 × 7.4 mm, respectively. Both testes were normal in size and echotexture. (a) Right testis. (b) Left testis. (c, d) Color Doppler ultrasonography showing normal blood flow in the testes. (c) Right testis. (d) Left testis.

In the present case, thermography revealed that the temperature of the right non-rugae scrotum was approximately 1°C higher than that of the left scrotum with rugae. Surprisingly, it was discovered that the temperature of the femoral skin adjacent to both sides of the scrotum was also elevated. Generally, the main factor in determining body surface temperature is skin blood flow, with arterial blood temperature usually higher than venous blood temperature. It appears that the high temperature in the femoral region may be influenced by peripheral arteries branching off from the femoral or internal iliac artery. Thus, it was speculated that the cooling function of the scrotum not only dissipates heat from the scrotal skin but also spatially separates the testes from the high-temperature femoral region.

As scrotal ultrasonography revealed no abnormal findings in the right testis in our patient with HSA without cryptorchidism, the patient's mother finally decided against scrotoplasty. Few reports indicate the relationship between the impaired testicular growth of normally descended testes and a high-temperature testicular environment. It has been reported in cultured testes that spermatogenesis is arrested before the early spermatocyte stage at 37°C.[13] Moreover, studies on *ex vivo* cultures of mouse testis explants reported that spermatogenesis fails to cross meiotic prophase I at 38°C, and some spermatocytes survive to the late pachytene stage but do not complete meiosis.[14] These results suggest that the testicular growth of the right testis of our patient with HSA may be impaired by high-temperature environments after puberty, especially during meiosis, even if the patient has not experienced impairment of testicular growth before puberty.

Conclusion

It was speculated that the cooling function of the scrotum is not only to dissipate heat from the scrotal skin, but also to spatially separate the testes from the high-temperature femoral region. Furthermore, it is necessary to consider the possibility that although testicular growth may not be impaired before puberty, it may get impaired during puberty due to the high-temperature environment if the testes are not protected by the scrotum's cooling function.

Author contributions

Hidenori Nishio: Conceptualization; data curation; writing – original draft. Kentaro Mizuno: Writing – review and editing. Daisuke Matsumoto: Data curation. Hideyuki Kamisawa: Writing – review and editing. Satoshi Kurokawa: Writing – review and editing. Akihiro Nakane: Writing – review and editing. Shoichiro Iwatsuki: Writing – review and editing. Tetsuji Maruyama: Supervision. Takahiro Yasui: Supervision. Yutaro Hayashi: Supervision.

Conflict of interest

Kentaro Mizuno is the Deputy Editor of *IJU Case Reports* and a coauthor of this article. He was excluded from editorial decision-making related to the acceptance and publication of this article. The other authors declare no conflict of interest.

Approval of the research protocol by an Institutional Reviewer Board

Not applicable.

Informed consent

Not applicable.

Registry and the Registration No. of the study/trial

Not applicable.

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