Acta Clin Croat **2026**, vol. **65**, no. **3**: 656-659

A Rare Case of Scrotal Fibrous Hamartoma of Infancy in Adult Male

Tena Piljušić[1], Goran Štimac[2], Matea Pirša[2], Šoip Šoipi[2], Monika Ulamec[3], Ivan Franin[3] and Borislav Spajić[2]

[1] Department of Urology, Dr Tomislav Bardek General Hospital, Koprivnica, Croatia;
[2] Department of Urology, Sestre milosrdnice University Hospital Center, Zagreb, Croatia;
[3] Ljudevit Jurak Department of Pathology, Sestre milosrdnice University Hospital Center, Zagreb, Croatia

CORRESPONDENCE TO Goran Štimac, MD, PhD, Department of Urology, Sestre milosrdnice University Hospital Center, Vinogradska cesta 29, HR-10000 Zagreb, Croatia
goran.stimac@icloud.com

RECEIVED September 13, 2024
ACCEPTED January 25, 2025

DOI 10.20471/acc.2026.65.03.17

SUMMARY

Fibrous hamartoma of infancy (FHI) is a rare, benign subcutaneous tumor occurring mainly before the age of 2 years. Typical localization is on the upper portions of the body, but FHI has also been known to appear in the genital region. It presents a diagnostic challenge due to localization and similarity to some malignant conditions. To our knowledge, no published article reports scrotal FHI in adulthood. Herein, we describe clinical presentation and management of a 44-year-old man with scrotal FHI.

KEYWORDS

Hamartoma; *Tumor*; *Benign lesions*; *Scrotal neoplasms*; *Diagnosis*; *Treatment*

Introduction

Fibrous hamartoma of infancy (FHI) is an exceedingly rare benign subcutaneous tumor that typically occurs in the first 2 years of life. No published article reports FHI in adulthood, though there is one case of recurrent lesion found at the age of 19. It typically occurs in the shoulder or axillary region of male infants. However, the genital area may also be affected. A distinct microscopic finding is that of three components, i.e., mature fat tissue, myofibroblastic component, and primitive mesenchymal cells[1,2,4-10,12,13]. We present a case of an adult 44-year-old man with FHI on the scrotum, with a focus on organ-sparing treatment.

Case report

A 44-year-old male patient was admitted to our clinic with a desire to remove a large verrucous skin mass on his left hemiscrotum which the patient stated was congenital. He was diagnosed and scheduled for surgery three years before but due to fear of the procedure, he postponed the surgery. The lesion was solitary and pigmented in the same manner as the surrounding scrotal skin, it was not painful but the patient reported decreased sensitivity in that area. Over years, the growth enlarged following the rest of the body (Fig. 1a). On palpation and ultrasound, both testicles were normal. The growth was not attached to underlying structures. Due to the age of the patient and the indolent course of the disease, no further diagnostics were made. At surgery, the tumor was completely excised and underlying intrascrotal structures were preserved.

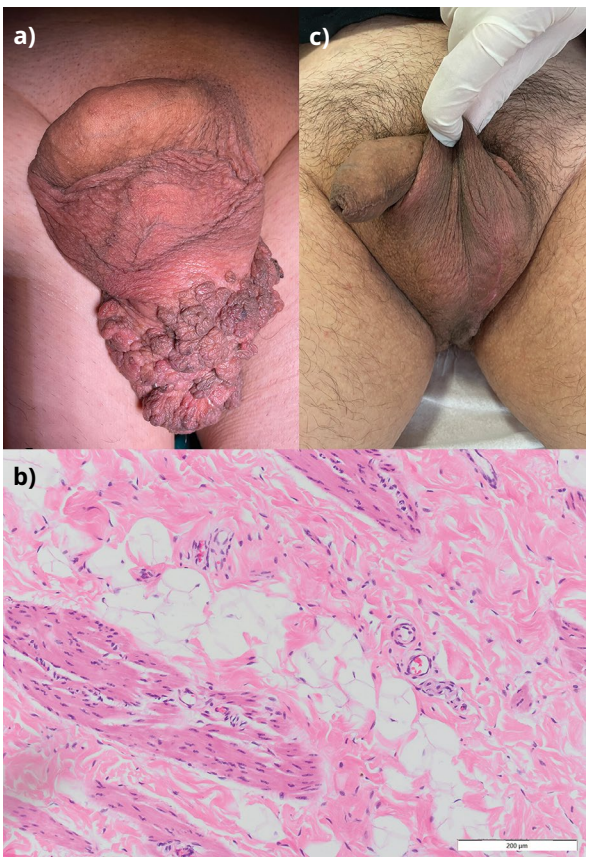


Fig. 1. (a) Preoperative presentation showing verrucous scrotal skin mass measuring 7×6 cm in size; (b) pathologic findings with all three components necessary for qualifying the lesion as fibrous hamartoma of infancy are present: mature fat tissue, myofibroblastic component, and primitive mesenchymal cells (40x, H&E); (c) postoperative finding with no recurrence and aesthetically pleasing result.

Gross inspection revealed a greyish verrucous tumorous lesion 7×6×3 cm in size, on a 3 cm diameter base. Microscopically, the surface featured verrucous, regular layers of the keratinized epidermis, with branches of nerves and blood vessels in hyalinized fibrous tissue, multiple smooth muscle fibers, and myofibroblasts, less mature adipose tissue and focal loose tissue with bluish foundation (Fig. 1c). Immunohistochemically the cells were positive for SMA, calponin, and desmin. CD34 was positive in blood vessel endothelium and S-100 was positive in nerve branches and adipose tissue. BCL-2 reaction was non-specific and p16 was negative.

Based on the available clinical information, typical histologic findings, and immunohistochemical analysis, the lesion was diagnosed as FHI with slow growth dynamics. At six-month follow-up after surgery, the patient experienced an uneventful recovery with no local recurrence and was satisfied with the cosmetic appearance of the scrotum after reconstruction (Fig. 1b).

Discussion

Whether FHI is a neoplastic process or local malformation is still unknown. Due to its peculiarity and rarity, only several investigators have reported cytogenetic aberrations, with no recurrent translocations or karyotypic abnormalities[2]. Organoid growth pattern justifies the term hamartoma[3].

Although FHI typically has an onset in the first 2 years of life, recent reports have extended the age range into later childhood. In addition to the usual location on the upper extremities, axilla, and upper back, the anatomic distribution has expanded to include almost any anatomic site[2,6].

Although reports on the occurrence in the inguinal and genital region are not uncommon and account for 17% of cases, there are only 26 reports of scrotal FHI in the literature[1-12]. Other locations in the genital region are the spermatic funicle, perineum, labia majora, and suprapubic region[5]. The congenital variant appears in approximately 20% of all cases and is usually recognized at birth or immediately afterward[8]. Usually, it is the child's parents who seek medical advice due to a growing lesion on the body. It predominantly occurs in males in a ratio of 2-3:1[2-9,12]. Our patient is the oldest patient currently reported in published English-language literature. Clinically, FHI is presented as a solid, painless, well-defined cutaneous or subcutaneous formation, with a tendency to grow. Usually, it is a small solitary lesion but multiple lesions are known to occur, and its size varies from 1 to 8 cm up to 17 cm[11]. On histologic examination, FHI has three characteristic components, i.e., well-differentiated bundles of fibrous connective tissue, primitive mesenchymal stroma arranged in a whirly pattern with numerous small capillaries, and irregular islands of mature adipose tissue[4-6,8,9,12]. FHI itself is poorly demarcated with diffused fibrous tendrils reaching in the neighboring soft tissue[5]. An unnecessary radical orchiectomy or superfluous cytotoxic therapy may occur if the diagnosis is questionable before surgery. Unless locally invasive or if an incomplete resection was performed, yearly ultrasonography with physical examinations may be a reasonable technique for follow-up as there are currently no practical guidelines for the management of FHI. Some authors advocate achieving clear surgical margins of at least 1 cm as known recurrences are speculated to be the result of a failure of complete resection, not achieving clear margins or destruction of tumor capsule and subsequent seeding of tumor cells in the surrounding tissue[1,8].

Conclusion

The period of FHI occurrence correlates with the most common tumors of the scrotum, which is why every solid formation in the scrotum with a tendency to grow deserves special attention. A triphasic histologic pattern with portions of each tissue varying among individual cases is the typical presentation of FHI. Although FHI itself is a rare entity, correct preoperative diagnosis with clinical and radiological findings can help avoid unnecessarily aggressive overtreatment[4,6,8,9,12]. Distinction in diagnosis between FHI and other possibly unfavorable scrotal lesions is worth making because FHI typically features a benign nature and local excision or complete resection is an appropriate choice of treatment. ■

References

1. Kim HK, Kim KS, Kang DW, Lee SY. Fibrous hamartoma of infancy in the scrotum: a case report. Korean Soc Radiol. 2017;76(2):152-7. doi: 10.3348/jksr.2017.76.2.152
2. Saab ST, McClain CM, Coffin CM. Fibrous hamartoma of infancy: a clinicopathologic analysis of 60 cases. Am J Surg Pathol. 2014;38:394-401. doi: 10.1097/PAS.0000000000000104
3. Harris CJ, Das S, Vogt PJ. Fibrous hamartoma of infancy in the scrotum. J Urol. 1982;127(4):781-2. doi: 10.1016/s0022-5347(17)54040-9
4. Stepančec H, Kokot Z, Keretić D, Radiković S, Grgurović D. Fibrous hamartoma of infancy in the scrotum. Eur J Pediatr Surg Rep. 2019;7:e100-e103. doi: 10.1055/s-0039-1697924
5. Popek EJ, Montgomery EA, Fourcroy JL. Fibrous hamartoma of infancy in the genital region: findings in 15 cases. J Urol. 1994;152(3):990-3. doi: 10.1016/s0022-5347(17)32639-3
6. Groisman G, Kerner H. A case of fibrous hamartoma of infancy in the scrotum including immunohistochemical findings. J Urol. 1990;144(2 Pt 1):340-1. doi: 10.1016/s0022-5347(17)39449-1
7. Stock JA, Niku DS, Packer GM, Krous H, Kaplan GW. Fibrous hamartoma of infancy: a report of two cases in the genital region. Urology. 1995;45(1):130-1. doi: 10.1016/s0090-4295(95)97410-5
8. Starr S, Mendhiratta N, Iskander PJ, Ye H, Sturm R. Scrotal fibrous hamartoma of infancy: a case report and literature review of a rare tumor of the genitourinary tract. Urol Case Rep. 2022 Sep 12;45:102218. doi: 10.1016/j.eucr.2022.102218
9. Al-Ibraheemi A, Martinez A, Weiss SW, Kozakewich HP, Perez-Atayde AR, Tran H. Fibrous hamartoma of infancy: a clinicopathologic study of 145 cases, including 2 with sarcomatous features. Mod Pathol. 2017;30: 474-85. doi: 10.1038/modpathol.2016.215
10. Dickey GE, Sotelo-Avila C. Fibrous hamartoma of infancy: current review. Pediatr Dev Pathol. 1999;2:236-43. doi: 10.1007/s100249900119
11. Qiao YJ, Yang WB, Chang YF, Zhang HQ, Yu XY, Zhou SH, Yang YY, Zhang LD. Fibrous hamartoma of infancy with bone destruction of the tibia: a case report. World J Clin Cases. 2022;10(30):11190-7. doi: 10.12998/wjcc.v10.i30.11190
12. Sotelo-Avila C, Bale PM. Subdermal fibrous hamartoma of infancy: pathology of 40 cases and differential diagnosis. Fetal Pediatr Pathol. 1994;14:39-52. doi: 10.3109/15513819409022024
13. Kapatia G, Chatterjee D, Gupta K, Trehan A. Fibrous hamartoma of infancy with sarcomatous transformation: an unusual morphology. Autops Case Rep [Internet]. 2022;12:e2021380. doi: 10.4322/acr.2021.380

SAŽETAK

Rijedak slučaj fibroznog hamartoma dječje dobi u odraslog muškarca

Tena Piljušić, Goran Štimac, Matea Pirša, Šoip Šoipi, Monika Ulamec, Ivan Franin i Borislav Spajić

Fibrozni hamartom dječje dobi (*fibrous hamartoma of infancy*, FHI) je rijedak dobroćudni tumor potkožnog tkiva koji se pojavljuje najčešće u djece prije navršene druge godine života. Tipično se pojavljuje u području gornjih ekstremiteta i ramenog obruča te rjeđe u genitalnoj regiji. Ponekad predstavlja dijagnostičku dilemu većinom zbog kliničke sličnosti s malignim tumorima genitalne regije. Koliko je nama poznato, dosad nije objavljen slučaj FHI skrotuma u odraslog muškarca. Naš cilj je prikazati dijagnostiku i liječenje 44-godišnjeg muškarca sa skrotalnim FHI.

KLJUČNE RIJEČI

Hamartom; *Tumor*; *Benigne lezije*; *Novotvorine skrotuma*; *Dijagnoza*; *Liječenje*