Fibrous hamartoma of infancy in the scrotum — Report of a case

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Abstract

Fibrous hamartoma of infancy (FHI) is rare benign tumors. FHI is typically present before the age of two years. We report a 13-month-old child with FHI, which presented as the growing scrotum mass clinically diagnosed rhabdomyosarcoma of the trunk. Right scrotal swelling was presented. The mass was irregular, solidly, and non-tender. Operative findings revealed a mass fixed to the scrotal fundus. FHI is differentiated from other malignant neoplasms in the scrotum.

1. Case report

A 13-month-old boy presented with right scrotal swelling noticed for one month. There were no evident congenital abnormalities at birth. There was no familial history of diseases.

Physical examination revealed a 3 × 3 cm growing, irregular, swelling, solitary and moveable mass with unclear borders in the right scrotum that was not tender when palpated, right testis was separated from the mass (Fig. 1).

Magnetic resonance imaging revealed a well encapsulated soft tissue mass lesion in the scrotum and separated from spermatic cord. The mass with poorly defined margin and a reticular pattern with an interposing fat component that shows a reduced signal on fat suppression inversion recovery image (Fig. 2). The
tumor might be one of malignant soft tissue tumors, most likely rhabdomyosarcoma.

At first, as the tumor was well encapsulated mass lesion in the scrotum and separated from testis (T) and spermatic cord, we used a transscrotal incision, and identified the mass, testicle and spermatic cord structures. Operative findings revealed that the testicle and spermatic cord structures were normal. The mass consisted of solid tissue measuring 3 cm and was separated with the tunica vaginalis (Fig. 3). The mass was fixed to the tunica dartos at the fundus of the scrotum. The mass was completely resected with preservation of the testicle and spermatic cord structures. Histopathological examination demonstrated mature adipose tissue, spindle-shaped fibroblastic cells in a collagenous stroma and immature round mesenchymal cells which was positive for CD34 immunohistochemically, suggesting this lesion is FIH (Fig. 4A–D). Follow up at 6 months revealed no evidence of recurrence.

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**Fig. 2.** MRI (T2) images obtained a well encapsulated soft tissue mass (M) lesion in the scrotum and separated from testis (T) and spermatic cord.

**Fig. 3.** Intraoperative findings revealed that the testicle and spermatic cord structures were normal. The mass consisted of solid tissue measuring 3 cm (arrow).

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**Fig. 4.** Histopathological findings revealed. (A) Dense mature fibro-collagen, (B) loose immature basophilic or mesenchymal tissue, (C) mature adipose tissue, which forms a vague, irregular organoid pattern, (D) CD34 immunohistochemistry shows reactivity in immature mesenchymal tissue, vascular endothelium.
2. Discussion

FHI tumors occur mainly on the armpit area, scapula and upper arm followed by the thigh, groin, buttocks, scrotum and the head/face. Almost all FHI tumors are solitary, moveable nodules with unclear borders and are located in the deep dermis or subcutaneous layer. Occasionally, the tumor involves the fascia or muscle surface, but does not generally involve the deep muscle.

The characteristic pattern of fibrous proliferation of this hamartoma occurring in the subcutaneous tissue was first reported by Reye in 1956 [2]. In 1965, Enzinger reported a review and named “fibrous hamartoma of infancy” based on its various clinical and histological features [3].

Clinically 20–25% of patients with FHI are recognized at birth and nearly all are diagnosed by 2 years of age [3,4]. The male to female frequency ratio is about 2:1 [1]. FHI is a benign tumor with no reported cases of metastasis, although rare cases of local recurrence have been reported following incomplete excision. Thus, optimal management requires complete excision [1,5].

In the current World Health Organization classification, FHI is defined as a poorly circumscribed superficial soft tissue mass with a characteristic organized pattern of 3 histological components: (1) dense fibrous connective tissue in well-defined bundles of that branch, weave, interweave, and project into fat. (2) Primitive mesenchyme arranged in nests, concentric whorls bands, rich in capillaries with a small distribution of lymphocytes. (3) Mature adipose tissue as islands that are intimacy admixed with the first two components [1]. The relative abundance of one kind of tissue may cause diagnostic error and confused with embryonal rhabdomyosarcoma [6]. Immature mesenchmal cell also expressed CD34 and SMA. Like this case, the presence of small vascular covered with CD34 positive endothelium indicated the tumors were well-vascularized [1]. CD34 shows reactivity in immature mesenchymal tissue, vascular endothelium.

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Conflict of interest
None.

References