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Benign intrascrotal lipoblastoma in a 4-month-old infant: a case report and review of literature

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Abstract Lipoblastomas are rare benign soft tissue tumors that occur primarily in young children. Most lipoblastomas occur in the extremities, trunk, head, and neck. An intrascrotal location is unusual. We describe a case of a 4-month-old infant with an intrascrotal lipoblastoma and discuss the differential diagnosis and review the literature.

Lipoblastoma is a very rare benign neoplasm of the fetal embryonal white fatty tissue occurring most commonly in infants and young children [1]. Boys are 3 times more commonly affected than girls [2]. This tumor presents as a localized well-circumscribed lesion (lipoblastoma) or as a multicentric lesion (lipoblastomatosis) [3]. Most lipoblastomas occur in the superficial tissues of the arms and legs but may also arise in the head and neck, parotid, eyelid, tonsillar fossa, trunk, mediastinum, and retroperitoneum [3]. We describe a case of a 4-month-old male child with an intrascrotal lipoblastoma. To the best of our knowledge, only 6 previous cases of lipoblastoma [1-6] have been reported in this rare site.

1. Case report

A 4-month-old male infant presented to the Pediatric Surgery Department, Al Hada Armed Forces Hospital, Kingdom of Saudi Arabia, with a huge, painless right-sided intrascrotal mass observed by the parents with a rapidly progressive course. On physical examination, there was a huge intrascrotal solid, soft, and painless mass more on the right side (Fig. 1), with both testicles palpated in the inguinal regions. Scrotal ultrasound and computed tomographic scan (Fig. 2) showed a large fatty tumor more at the right side and bilateral inguinal testes. The lesion was approached through a right inguinal incision, and the fatty scrotal tumor was easily dissected from the dartos muscle, except at the right side where it was tightly adherent to the scrotal layers. The tumor was totally removed. The testes were spared and had been displaced by this large tumor to the inguinal region. Both testes were surgically fixed to the scrotum. On macroscopic appearance, the tumor was solid, was encapsulated, appeared grayish-yellow, and measured 10 × 9 × 6 cm (Fig. 3). The cut surface showed a lobular arrangement. Histologic examination of the mass (Figs. 4 and 5) showed white fat with lobular architecture separated by strands of fibrous septae of varying degrees of differentiation. Most adipocytes were mature and interspersed with various types of lipoblasts. Some of the lipoblasts were small and

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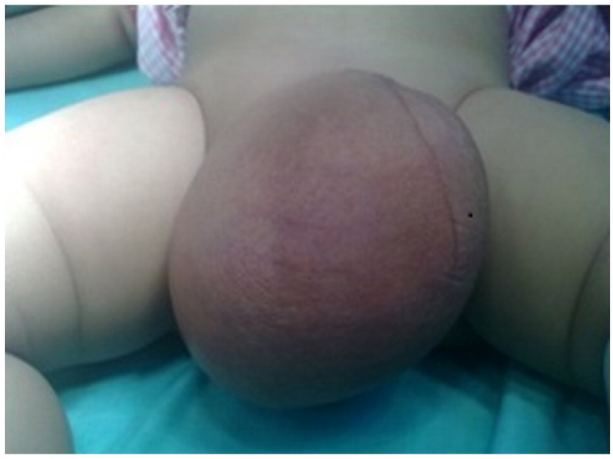

Fig. 1 Four-month-old male infant with a large disfiguring scrotal mass.

round with a signet ring appearance; others were larger and had a multivacuolated cytoplasm as well as a notched nucleus. Poorly differentiated mesenchymal cells (stellate and nonvacuolated spindle cells) were also present and embedded in a myxoid stroma. There were no mitotic figures. A cytogenetic analysis was not available because of lack of fresh tumor tissue. The patient had an uncomplicated recovery and was free of disease at 13 months postoperative follow-up.

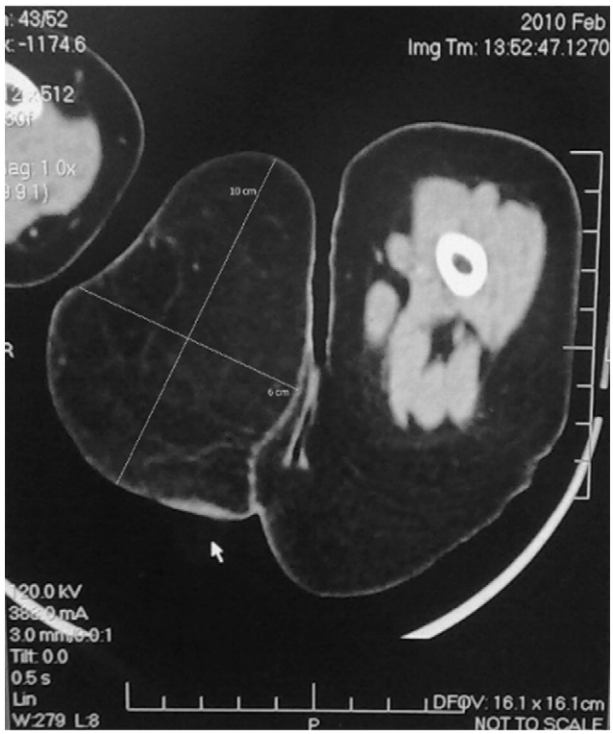

Fig. 2 Computed tomographic scan of the mass showing a large fatty scrotal tumor (10 × 6 cm).

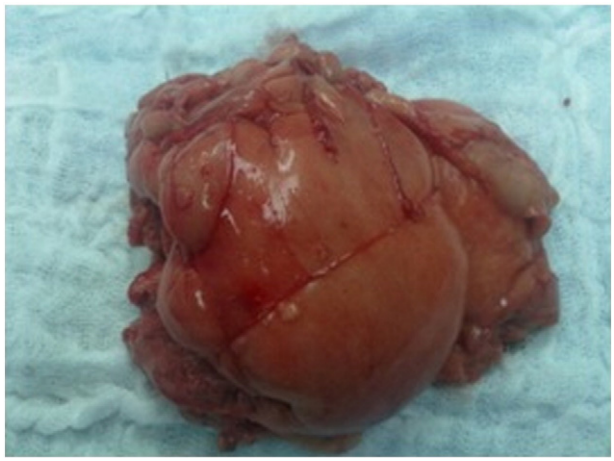

Fig. 3 Gross appearance of the resected specimen. It is 10 × 9 × 7 cm in diameter with grayish-yellow color and lobular architecture.

2. Discussion

Lipoblastoma and lipoblastomatosis are rare benign mesenchymal tumors of fetal white fat tissue that are almost exclusively observed in young children. Only rare examples have been reported in older children and young adults [7]. Lipoblastoma has also been referred to as embryonal lipoma, benign childhood adipocytic tumor, fetal lipoma, and infantile lipoma [8]. In the pediatric population, adipose tumors represent approximately 6% of all soft tissue neoplasms. Approximately two thirds of these are simple lipomas or variants, whereas up to 30% are lipoblastomas [9]. Van Meurs [10] in 1947 was the first to describe the ability of a lipoblastoma to differentiate into a common lipoma. This observation supports the concept that lipoblastomas are a result of a

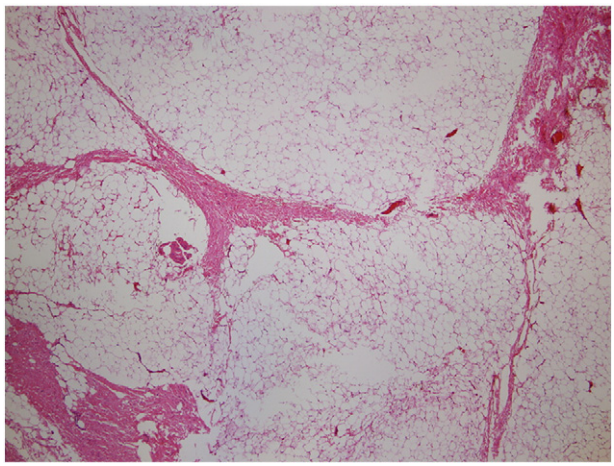

Fig. 4 Microscopic view of the tumor showing white fat with lobular architecture separated by strands of fibrous septae and areas of myxoid changes (hematoxylin and eosin stain, original magnification ×40).

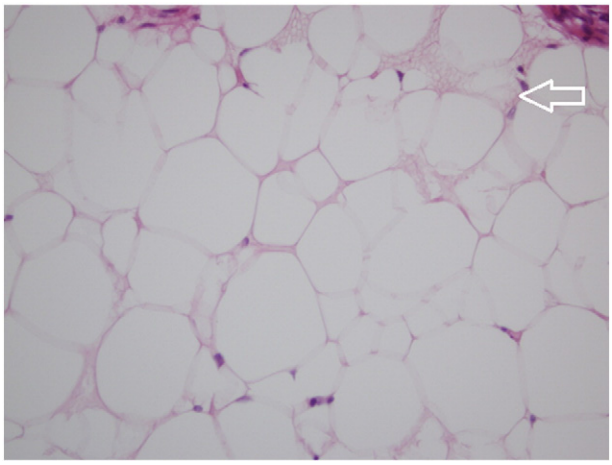

Fig. 5 Higher magnification of this tumor demonstrating cell with pale multivacuolated cytoplasm with nuclear indentation (lipoblast; arrow), suggesting lipoblastoma (hematoxylin and eosin stain, original magnification ×400).

continued proliferation of immature fat cells and lobules in the postnatal period. Lipoblastoma and lipoblastomatosis are generally benign, and the treatment of choice for these is surgical excision [2]. Local recurrence may occur, particularly with lipoblastomatosis, when the excision is incomplete [3]. Metastasis has not been described [3]. To our knowledge, only 6 cases of lipoblastomas have been previously reported in the scrotum in the English literature, all were painless and in none was the diagnosis suspected before surgery [1-6]. In our patient, lipoblastoma was listed among the probable conditions in the differential diagnoses before surgery. After resection of the mass, the diagnosis of lipoblastoma was confirmed by histopathologic studies. Table 1 summarizes the clinical data of the previously reported cases and the present case. The age of the patients in these cases ranged from 7 to 48 months; the diameter of the tumors varied from 2.3 to 14 cm, and in 1 case, the mass extended to the perineum [1]. Our patient is the youngest reported case, and the mass was the second largest tumor after that reported by Turner et al [4].

All cases were treated by local excision, and the ipsilateral testis was separated from the mass and spared in 5 cases (including the present case).

The differential diagnosis may be very difficult because of the close resemblance between lipoblastomas and myxoid or well-differentiated liposarcomas and hibernomas [2,3]. The young age of the patient, the location, the prominent lobulation, and the lack of either atypical nuclei or abnormal mitoses suggest the diagnosis of lipoblastoma [11]. Microcystic spaces filled with mucin can be seen in both lipoblastoma and liposarcoma, although some authors consider them as a hallmark of liposarcoma. The zoning phenomenon (immature mesenchymal cells and myxoid stroma more prominent at the periphery of the lobules) is reported as being typical of lipoblastoma but is not always present [3]. Hibernoma must also be considered in the differential diagnosis. However, this is a soft tissue tumor of brown fat and has a characteristic gray-brown, lobulated structure. The fat cells have more intracytoplasmic vacuoles as compared with lipoblastoma, and the appearance is granular [12]. Recent studies have described specific chromosomal abnormalities that characterize benign and malignant adipose tissue tumors. The specific clonal chromosomal alteration, t (12;16)(q13;p11), is characteristic of myxoid liposarcomas. Lipoblastomas typically have chromosomal abnormalities involving band 8q12, resulting in the rearrangement of the pleomorphic adenoma gene 1 oncogene, but some only have polysomy for chromosome 8 [13]. Therefore, the tumor karyotype may help pathologists distinguish between lipoblastomas and myxoid liposarcomas in most cases. However, surgical excision is always the mandatory.

Benign lipoblastoma and lipoblastomatosis are rare benign tumors of fetal embryonal white fatty tissue affecting exclusively infants and children. They may cause significant difficulty in differential diagnosis for the pathologist because of close resemblance to that of myxoid or well-differentiated liposarcoma and hibernoma. Surgical excision is the treatment of choice.

Table 1 Summary of the clinical data of the previously reported cases of pediatric scrotal lipoblastomas (including our case)

Reference	Age (mo)	Site	Perineum involvement	Size (cm)	Testes	Follow-up time (mo)	Recurrence
Arda et al [1]	15	Left scrotum	Yes	6.5 × 4 × 3, 45 g	Spared	10	No
Chun et al [5]	9	Left scrotum	No	5 × 2.5 × 1.5	Not mentioned	12	Not mentioned
Somers et al [6]	18	Not mentioned	No	2.3 × 2 × 1.3	Not mentioned	48	Not mentioned
Turner et al [4]	7	Left scrotum	No	14 × 14 × 8, 314 grams	Spared	6	Yes, 2 scrotal lipoblastomas after 6 months
Del Sordo et al [3]	48	Right scrotum	No	2 × 1.2 × 0.8	Spared	4	No
Dy et al [2]	48	Not mentioned	No	3.5 × 2	Spared	Not mentioned	Not mentioned
Present case	4	Right scrotum	No	10 × 9 × 7	Spared	12	No

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