

# Primary Inguinal Lymphangioma, Cryptorchidism and Inguinal Hernia Co-occurrence in a Child: Case Report

## Bir Çocukta Primer İnguinal Lenfanjiom, İnmemiş Testis ve Kasık Fıtığı Birlikteliği

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**ABSTRACT** Cryptorchidism, inguinal hernia and hydrocele are the most common pathologies of the inguinal region. Unlike these common pathologies, there may be some unusual pathologies in the inguinal region such as neurofibromatosis, lymphangioma, neuroblastoma and dermoid cyst. A nine-month old boy who presented with a right groin soft mass of 5x6cm beginning from the border of external inguinal ring to the base of the penis together with right inguinal hernia and right non-palpable testis was operated and histopathological examination revealed benign vascular proliferation which is compatible with lymphangioma. The importance of this case is that the lymphangioma was primarily located in the inguinal region, there was co-occurrence of lymphangioma, cryptorchidism and inguinal hernia and there is a possibility of the lesion causing cryptorchidism because of mass effect.

**Key Words:** Lymphangioma; cryptorchidism; hernia, inguinal; child

**ÖZET** İnmemiş testis, kasık fıtığı ve hidrosel inguinal bölgenin en sık rastlanan patolojileridir. Bunların dışında nörofibromatozis, lenfanjiom, nöroblastom, dermoid kist gibi daha az görülen patolojilere de bu bölgede rastlanılabilir. Sağ kasık bölgesinde 5x6 cm boyutlarında, dış inguinal halka hizasında başlayıp penis köküne uzanan yumuşak kitlesi olan ve buna eşlik eden sağ palpe edilemeyen testis, sağ kasık fıtığı saptanan dokuz aylık erkek çocuk ameliyat edildi. Histopatolojik incelemesi lenfanjiom ile uyumlu benign vasküler proliferasyon şeklinde geldi. Bu olgunun önemi, lenfanjiomun primer olarak inguinal bölgede yer alması, lenfanjiom ile inmemiş testis ve kasık fıtığının birliktelik göstermesi ve lezyonun kitle etkisinden dolayı inmemiş testise neden olmasıdır.

**Anahtar Kelimeler:** Lenfanjiyoma; kriptorşidizm; fıtık, inguinal; çocuk

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Cryptorchidism, scrotal or abdominoscrotal hydrocele and inguinal hernia are the most common pathologies of the inguinal region. Unlike these, there may be some unusual lesions in the inguinal region such as neurofibromatosis, lymphangioma, neuroblastoma and dermoid cyst.<sup>1-4</sup>

According to our literature survey, inguinal cystic lymphangioma is a very rare entity and synchronous presentation of inguinal hernia, cryptorchidism and inguinal cystic lymphangioma has not been reported previously. The aim of this study is to present an infant who was operated

because of bilateral cryptorchidism, bilateral inguinal hernia and right inguinal cystic lymphangioma which was diagnosed prenatally in fetal magnetic resonance imaging (MRI).

## CASE REPORT

A nine-month old boy presented with a right groin soft mass of 5x6cm extending from the border of external inguinal ring to the base of the penis together with right inguinal hernia and right non-palpable testis (Figure 1). A fetal MRI revealed multilocular mass of 49x38 mm in the scrotal region (Figure 2) and MRI at 2-month old revealed a large multi-septated cystic lesion in the right inguinoscrotal region with enhancing thin wall and septations (Figure 3). The infant had no predisposing factors such as prematurity. He had underwent emergency surgery in another center when he was two-months old for left incarcerated hernia and left cryptorchidism. The child was admitted to our hospital when he was nine months old with groin mass. Inguinoscrotal ultrasonography (USG) revealed right canalicular testis whose dimensions are similar to contralesional testis, inguinal hernia and multicystic lesions at the right site with a preliminary diagnosis of lymphangioma. At surgery, the inguinal region was found to be covered with multicystic lesion from external inguinal ring to the base of penis and anterolateral border of the bladder. The testis was found to be in-

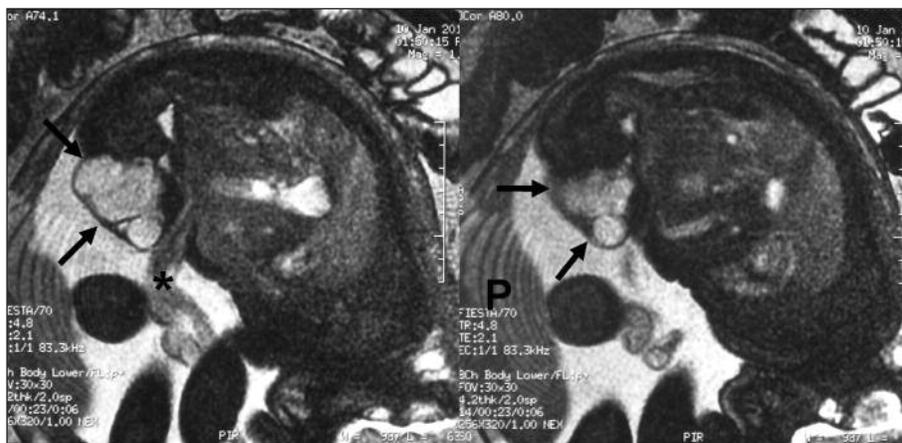
traabdominal located at the internal inguinal ring associated with a huge hernia sac (Figure 4). The lesion was excised and a high ligation of the hernia sac as well as orchidopexy of the testis, were performed. There were no intraoperative or postoperative complications. Histopathological examination revealed benign vascular proliferation compatible with lymphangioma.

## DISCUSSION

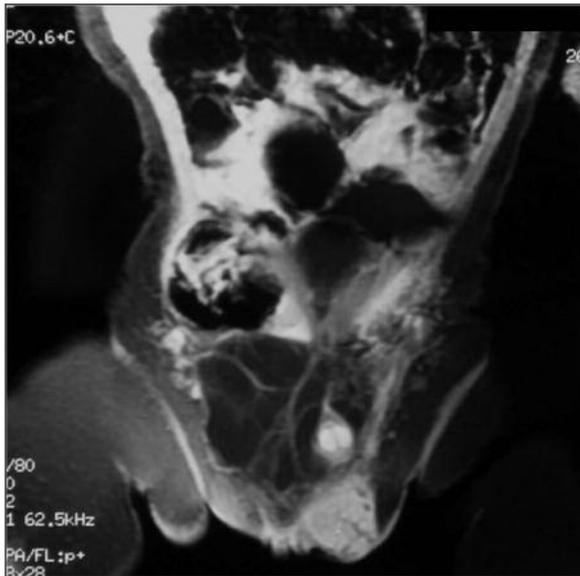
This case is the first presented case with cooccurrence of cryptorchidism, inguinal hernia and cystic lymphangioma in the inguinal canal.



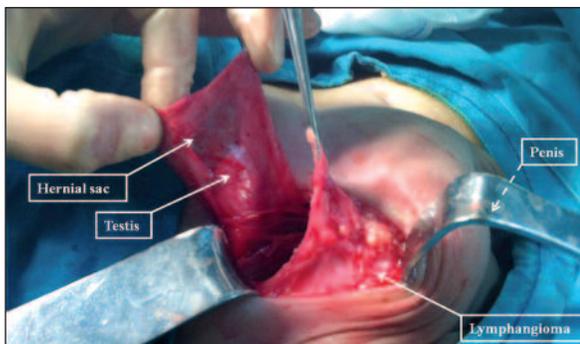
**FIGURE 1:** Right groin soft mass of 5x6 cm.  
(See color figure at <http://pediatri.turkiyeklinikleri.com/>)



**FIGURE 2:** Contiguous T2-weighted images of the fetus in the sagittal plane of the baby demonstrates the multilocular mass in the scrotal region (arrows). Asterisks indicates the umbilical cord and P is placenta.



**FIGURE 3:** Contrast-enhanced coronal T1-weighted image reveals a large multi-septated cystic lesion in the right inguinoscrotal region encircling the base of penis with enhancing thin wall and septations.



**FIGURE 4:** Intraabdominal testis, inguinal hernia with huge sac and primary inguinal lymphangioma.

(See color figure at <http://pediatri.turkiyeklinikleri.com/>)

The most common inguinal region pathologies are inguinal hernia and cryptorchidism with the incidence of 1-5% and 3-5% in full-term male newborns respectively. These pathologies may be seen synchronously.<sup>5,6</sup>

Although these pathologies are the most common pathologies of this region, there may be other unusual reasons of inguinal masses which have very rare occurrences such as neuroblastoma, lymphangioma, dermoid cyst, neurofibroma, liposarcoma and hemangioma.<sup>1-4</sup>

Lymphangioma is a benign tumor which is caused by congenital malformation of the lymphatic system.<sup>7</sup> Besides, they may also occur secondary to trauma.<sup>8</sup> Inguinal presentation is the least frequent site of lymphangiomas.

Although the literature survey revealed that inguinal lymphangiomas are the extension of retroperitoneal or intraabdominal the upper border of the lymphangioma was beginning from the external ring and extending to the base of the penis in our case.<sup>9,10</sup> Primary inguinal lymphangioma has not been reported previously in the literature.

USG and MRI are useful for preoperative diagnosis and to determine the extent of tumor.<sup>7</sup> Sonographic appearance of cystic mass with anechoic fluid is characteristic.<sup>9</sup>

Management options of lymphangiomas include observation, drainage, sclerotherapy, laser therapy and surgical excision.<sup>11</sup> The gold standard treatment of cystic lymphangiomas is surgical excision, when possible. Surgical excision is especially indicated when there is an increase in the cyst size.<sup>12</sup>

The importance of this case is that the lymphangioma was primarily located in the inguinal region, there was cooccurrence of lymphangioma, cryptorchidism and inguinal hernia and there is a possibility of the lesion causing cryptorchidism because of mass effect.

Although this case had a prenatal diagnosis, he had to be operated in another center in emergent conditions for incarcerated hernia. It would be better if this case would be followed in one center since birth however he admitted to our hospital when he was nine months old.

The etiology of right cryptorchidism in this patient may be the misadherence of the gubernaculum. Although this patient had left cryptorchidism, right cryptorchidism may be associated with the mass effect of lymphangioma by preventing the inguinal phase of testicular descent which was similarly discussed by Soyer et al.<sup>1</sup> The fate of the right testis could be the scro-

tum however because of the presence of lymphatic malformation in the inguinal region during the critical time of fetal descent of the testis, testicular descent was arrested and the testis remained in the abdomen. The left testis had descended until distal of inguinal canal therefore although it is difficult to prove the reason of intraabdominal location of right testis may be speculated as the mass effect of the lymphangioma.

## CONCLUSION

Although co-occurrence of cryptorchidism, inguinal hernia and lymphangioma is a very rare entity, pediatric surgeons should keep in mind that unusual masses can be seen in inguinal region which may cause cryptorchidism because of mass effect during antenatal period by preventing descent of ipsilateral testis.

## REFERENCES

1. Soyer T, Vargel I, Ayva S, Cavuşoğlu T, Cesur O, Bülbül S, et al. Intrasrotal extratesticular neurofibroma as a possible cause of failed descent in ipsilateral testis. *Indian J Pediatr* 2012;79(1):117-9.
2. Poenaru D, Jacobs DA, Kamal I. Unusual findings in the inguinal canal: a report of four cases. *Pediatr Surg Int* 1999;15(7):515-6.
3. Abantanga FA. Groin and scrotal swellings in children aged 5 years and below: a review of 535 cases. *Pediatr Surg Int* 2003;19(6):446-50.
4. Leeming R, Olsen M, Ponsky JL. Inguinal dermoid cyst presenting as an incarcerated inguinal hernia. *J Pediatr Surg* 1992;27(1):117-8.
5. Mattei P. Inguinal hernia and hydrocele. *Fundamentals of Pediatric Surgery*. 1<sup>st</sup> ed. New York: Springer; 2011. p.663-72.
6. Mattei P. Undescended testis. *Fundamentals of Pediatric Surgery*. 1<sup>st</sup> ed. New York: Springer; 2011. p. 673-8.
7. Vikićević J, Milobratović D, Vukadinović V, Golubović Z, Krstić Z. Lymphangioma scroti. *Pediatr Dermatol* 2007;24(6):654-6.
8. Goel R, Agarwal A, Lomanto D. Large lymphangioma presenting like irreducible inguinal hernia: a rare presentation and literature review. *Ann Acad Med Singapore* 2011;40(11): 518-9.
9. Popli MB, Garg DP, Anand R, Jain M, Jain N, Mahajan R. Retroperitoneal lymphangioma presenting as an inguinal hernia. *Indian Pediatr* 1994;31(12):1573-5.
10. Chiari G, Verga G. [Cystic lymphangioma of the greater omentum appearing as an inguinal hernia in an 18-month-old child]. *Chir Pediatr* 1989;30(6):290-1.
11. Solari V, Mullassery D, Lansdale N, Jesudason EC. Laparoscopic excision of a retroperitoneal lymphatic malformation in a newborn. *J Pediatr Surg* 2011;46(2):e15-7.
12. Uba AF, Chirdan LB. Management of cystic lymphangioma in children: experience in Jos, Nigeria. *Pediatr Surg Int* 2006;22(4):353-6.