

Lymphangiocoele: A very rare cause of primary infertility

Sunita Samal,
Sudeepta Kumar Swain¹,
P. Pallavee,
Seetesh Ghose
 Departments of Obstetrics
 and Gynecology and ¹General
 Surgery, MGMC and RI,
 Puducherry, India

Address for correspondence:

Dr. Sudeepta Kumar Swain,
 G-A Type 1 Quarters, MGMC
 and RI, Pilliyarkuppam,
 Pondicherry - 607 402, India.
 E-mail: sudeeptapappu@
 rediffmail.com

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ABSTRACT

There are limited causes of surgically treatable male infertility. Lymphangiocoele of scrotum is a very rare condition particularly in adult life. Lymphangiocoele causing infertility is further rare and not reported in English literature so far. We report an extremely rare case of lymphangiocoele in a 29 years male presenting with male infertility that improved after surgery.

KEY WORDS: Lymphangiocoele, lymphangioma scrotum, male infertility

INTRODUCTION

Primary infertility can be due to the male partner that should be investigated in all cases. Surgically treatable causes of male infertility are limited out of which varicocele is the commonest. There are some controversies regarding the surgical outcome of varicocelectomy on the improvement of fertility. But, literature from various meta-analyses suggests surgery as the first-line treatment in symptomatic cases with unexplained infertility. Lymphangiocoele of scrotum is a condition that is very difficult to differentiate from varicocele. As such lymphangiocoele of scrotum is very rare in adulthood, lymphangiocoele causing infertility is extremely rare and has not been described in English literature. Here, we report a case of 29 years male with lymphangiocoele who presented with oligospermia and primary infertility whose sperm motility and count improved after surgery.

were normal. On examination of the genitalia, he had grade 3 bilateral varicocele that was associated with intermittent dull aching pain. Ultrasonography of scrotum was equivocal for varicocele. Ultrasound abdomen did not reveal any abnormality. After counseling, bilateral varicocelectomy, was planned with trans-scrotal approach. Intra-operatively there was bilateral lymphangiocoele extending up to deep inguinal ring [Figures 1 and 2]. Bilateral testicular volume appeared normal with normal pampiniform plexus of veins. All the dilated tortuous lymphatics were excised up to deep ring leaving testicular artery and venous plexus intact. Postoperative recovery was uneventful, and he was discharged on 2nd postoperative day. There was no recurrence on follow-up till 9 months and improvement in seminal parameters (sperm count 23 million/ml, progressive motility 38%) had been noted. At present, the couple is blessed with 12 weeks of pregnancy.

CASE REPORT

A 29 years male presented to our hospital with 5 years of primary infertility. Female partner had been investigated extensively and was found to be normal. His seminal analysis revealed oligospermia (9.2 million/ml) and poor progressive motility (22%). Other parameters were normal. He was referred to surgical outpatient department for further evaluation. His secondary sexual characters

DISCUSSION

Lymphangiocoele of scrotum is sparsely described in literature as it is a rare entity. There is some confusion to differentiate it from lymphangioma of scrotum. Lymphangioma of scrotum is usually a congenital disorder causing malformation of part of lymphatics draining the scrotum and testis that also is rare as compared to the incidence of congenital lymphangioma of other parts of

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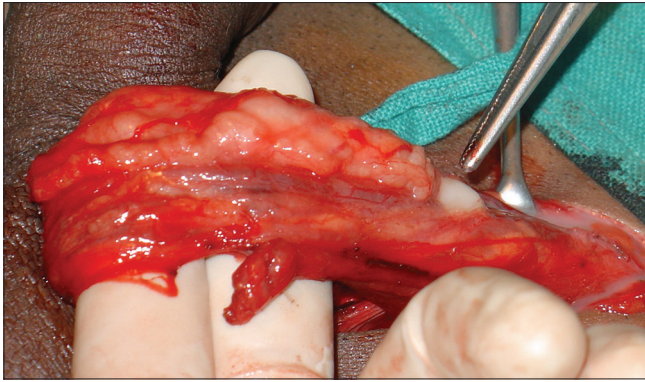


Figure 1: Lymphangiocoele around spermatic cord

the body.^[1,2] Usual presentation is in childhood presenting as painless localized swelling of the scrotum. It may be associated with lymphangioma of other area, particularly of perineum and thigh.^[3] Whereas lymphangiocoele involves the lymphatics of scrotum and spermatic cord like varicocele and clinically presents with physical findings similar to varicocele. Lymphangioma of scrotum presenting in adulthood is very rare.^[4] Lymphangiocoele is almost indistinguishable from varicocele on clinical examination. Ultrasonography should be able to differentiate it from varicocele. But, sometimes it is inconclusive like in our case. Like varicocele, it can contribute to infertility as theoretically it produces the same effect on spermatogenesis as varicocele.^[5] Since the unavailability of good literature, the treatment plan and indication of surgery may be similar to varicocele. This is probably the first case in literature where bilateral lymphangiocoele presented with male infertility. Postoperative improvement in seminal parameters and proven fertility confirms lymphangiocoele as the etiology for male infertility.

CONCLUSION

Lymphangiocoele is a very rare disorder that can present in adulthood and may contribute to male infertility. Surgical

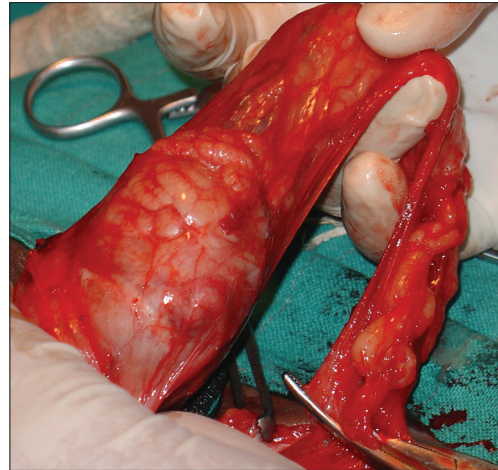


Figure 2: Lymphangiocoele around testis

treatment can be considered as an option to treat infertility in these cases. However with limited literature, it is difficult to describe this condition and recommend exact treatment algorithm at present.

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