MICROSURGICAL VARICOCELECTOMY IN PRE-PUBERTAL CHILDREN: SAFETY, EFFICACY, AND REPRODUCTIVE IMPLICATIONS - A CASE REPORT AND LITERATURE REVIEW

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ABSTRACT

Objective: *In this report, we aim to present a case of a prepubertal child with left varicocele who underwent successful microsurgical ligation.* ***Case(s) Presentation:*** *A 9-year-old boy was admitted to urology clinic due to his parents complaint about a mass in his left scrotum. We diagnosed this patient with grade III varicocele (Dubin-Amelar classification). The patient underwent microsurgical ligation surgery. After surgery, the mass such as "bag of worms" disappeared and the testicular volume increased by 53% from preoperative volume (1.8 to 2.3 cc).* ***Discussion:*** *The results of this case report indicate that microsurgical ligation of varicocele in children is a safe and effective therapeutic option.* ***Conclusion:*** *Microsurgical ligation of varicocele in pre-pubertal children represents a safe and effective therapeutic option, offering symptom relief and potential preservation of future fertility. Further studies are warranted to elucidate the long-term outcomes and reproductive implications of microsurgical varicocelectomy in this population.*

Keywords:*Microsurgical ligation, varicocele, children.*

ABSTRAK

Tujuan: *Laporan ini bertujuan untuk menyajikan kasus seorang anak prapubertas dengan varikokel kiri yang berhasil menjalani ligasi mikrosurgis.****Presentasi Kasus:*** *Seorang anak laki-laki berusia 9 tahun dirawat di klinik urologi karena orang tuanya mengeluhkan adanya massa di skrotum kirinya. Kami mendiagnosis pasien ini dengan varikokel tingkat III (klasifikasi Dubin-Amelar). Pasien menjalani operasi ligasi mikrosurgis. Setelah operasi, massa seperti "kantong cacing" menghilang dan volume testis meningkat sebesar 53% dari volume sebelum operasi (1,8 hingga 2,3 cc).* ***Diskusi:*** *Hasil laporan kasus ini menunjukkan bahwa ligasi mikrosurgis varikokel pada anak-anak merupakan pilihan terapi yang aman dan efektif.* ***Simpulan:*** *Ligasi mikrosurgis varikokel pada anak prapubertas merupakan pilihan terapi yang aman dan efektif, menawarkan peredaan gejala dan potensi pelestarian kesuburan di masa mendatang. Penelitian lebih lanjut diperlukan untuk menjelaskan hasil jangka panjang dan implikasi reproduksi dari varikokelektomi mikrosurgis pada populasi ini.*

Kata kunci: *Ligasi mikrosurgis, varikokel, anak.*

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INTRODUCTION

A varicocele develops due to an anomalous expansion of the pampiniform venous plexus, which is responsible for draining blood from the testicles to the gonadal vein trunci, as well as the pudendal and cremasteric veins. Varicoceles are predominantly seen on the left side.[1] Pediatric varicocele has an overall prevalence of 15%, being uncommon in boys under 10 years of age and becoming more frequent as puberty begins. Varicocele develops during accelerated body growth and increased blood flow to the testes, by a mechanism that is poorly understood. Genetic factors may also play a role.[2] It can damage testicular function, interfering with Sertoli cell proliferation, hormone secretion, testicular growth, and spermatogenesis. Left testicular volume loss is found in 70% of patients with grade II and III varicocele. Abnormal reproductive hormonal levels (increased serum levels of FSH and LH, and

decreased levels of inhibin B) and semen quality have been reported in varicocele patients, directly related to varicocele severity.[3]

Several studies have suggested that early correction of varicocele could prevent the decline in fertility found among men with varicocele discovered in adolescence. However, there is currently no evidence that treating varicocele in children will offer better andrological outcomes than performing the operation later[2]. Recommended indication criteria for varicocele correction in children and adolescents include symptomatic varicocele, where pain is present, and reduced total testicular volume (left + right) compared to normal testes, which is a promising indication criteria.[3]

Once the decision has been made to correct a varicocele in an adolescent or child, the chosen method becomes an important consideration. The best repair technique for correcting pediatric or adolescent varicocele is still debated. Several treatment options are available, including surgical (e.g., open inguinal-Ivanissevich, high retro-peritoneal-Palomo, subinguinal, high inguinal, microsurgical-inguinal and subinguinal, laparoscopic) and radiological (sclerotherapy, embolization, antegrade vs. retrograde) approaches.[4] The most frequent complications associated with varicocele repair include hydroceles, testicular atrophy, and recurrence. In this report, we aim to present a case of a prepubertal child with left varicocele who underwent successful microsurgical ligation.

CASE(S) PRESENTATION

Patient Information: A 9-year-old boy was admitted to our urology outpatient clinic due to his parents' complaint about a mass resembling a "bag of worms" in his left scrotum. The patient denied experiencing any discomfort or swelling in the scrotum or stomach during daily activities. The patient underwent circumcision at the age of four but has not yet shown signs of puberty. He has no prior history of genital issues, particularly scrotal problems, and there were no hematological or birth anomalies identified in his medical history. Additionally, there is no record of any previous surgical procedures. However, his father has been diagnosed with varicocele, identified three years after marriage, during which they had not yet conceived.

Physical examination results revealed normal physiological functions, including a pulse rate of 90 beats per minute, blood pressure of 100/70 mmHg, respiration rate of 22 breaths per minute, temperature of 36.0°C, height of 128 cm, and weight of 30 kg. Abdominal examination showed no palpable mass. Local examination of the scrotum revealed a mass resembling a "bag of worms" in the left scrotum while standing (Figure 1), and the Valsalva maneuver was positive. Laboratory tests, including routine blood tests and urinalysis, showed normal findings, and serum levels of FSH and LH were within normal ranges. Considering the patient's age and with the consent of the parents, semen analysis was not performed.

Figure1. Showinga“bagofwarm”inleftscrotum.

Image Findings: Scrotal ultrasound imaging revealed that the volume of the right testis was 2.14 cc, with normal epididymis and vascularization. The diameter of the right pampiniform plexus in the supine position was 1.7 mm, and the Valsalva maneuver was negative. However, the volume of the left testis was 1.8 cc (16% smaller than the right testicular volume), also with normal epididymis and vascularization. The diameter of the left pampiniform plexus in the supine position was 2.8 mm (65% wider than the right pampiniform plexus), and there was reflux of 3.3 mm during the Valsalva maneuver according to color Doppler ultrasound (Figure 2). Based on Sarteschi's criteria, the ultrasound imaging of the left scrotum revealed grade four varicocele. Additionally, an abdominal ultrasound and CT scan of the abdomen showed normal kidneys and no intra-abdominal mass.

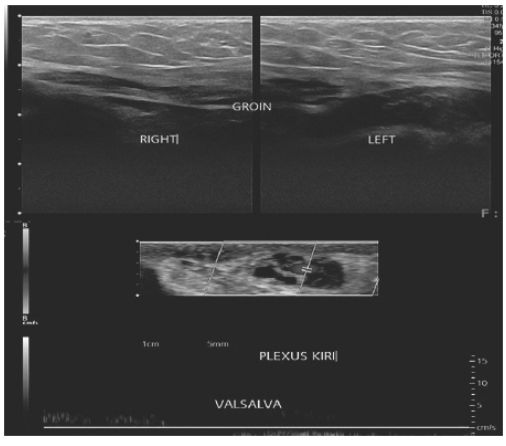

Figure 2. Colour doppler ultrasound showing dilatated plexus pampiniform while valsava maneuver

Figure 3. Dilatated plexus pampiniform (intra-operative finding).

Diagnostic Assessment: Based on physical examination and imaging findings, the patient was diagnosed with grade III varicocele according to the Dubin-Amelar classification. Therapeutic Intervention: Subsequently, the patient underwent microsurgical ligation surgery with optical magnification, employing an artery-sparing and lymphatic-sparing varicocelectomy (Figure 3)

Follow-up and Outcomes: The patient was in good condition after surgery and was discharged on the third day following the intervention. At the one-week follow-up, there were no signs of hydrocele or other signs of complications (Figure 4). Continued follow-up two months post-surgery revealed a significant increase in the volume of the left testis from 1.8 cc to 2.3 cc (Figure 5).

Figure 4. Follow up one month after surgery.

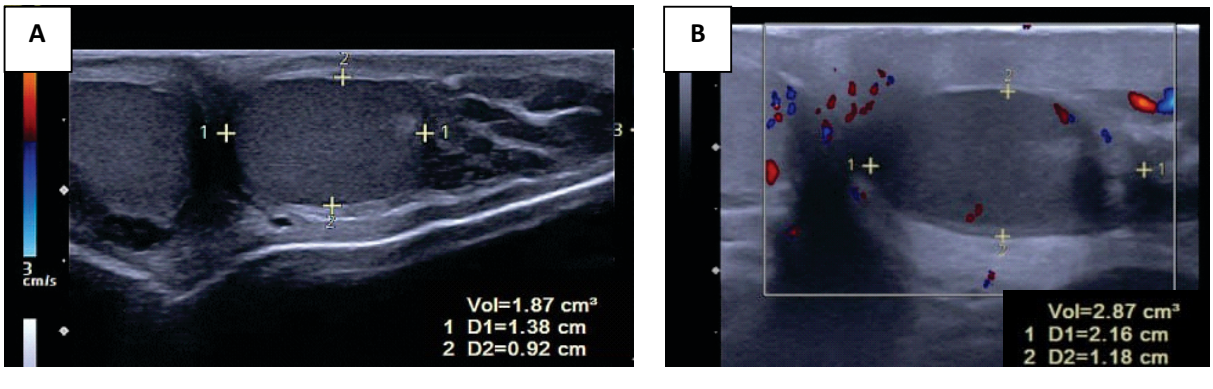

Figure 5. Left testicular ultrasound (A) Before surgery, testicular volume was 1.87 cc; (B) 2 months After surgery, testicular volume increased to 2.87 cc.

Patient Perspective and Informed Consent: The patient and his parents were satisfied and patient complaints have improved after the surgery. The patient's parent provided consent for the clinical data to be published, and all presented data have been anonymized to ensure patient confidentiality.

DISCUSSION

Varicoceles are rare in boys younger than 10 years old[1]. Although often showing no symptoms, varicoceles are usually detected by the patient, caregivers, or identified by paediatrician during a check-up.[2] In this case, a 9-year-old was admitted to urology clinic due to his parents has found the mass feels like a "bag of worms" in the left scrotum. The term "bag of worms" in scrotum refers to varicocele, which is characterized by abnormal dilatation and enlargement the pampiniform venous plexus.[5] By the anatomy, the veins in the testicle come together to make the pampiniform plexus. From there, the blood travels up through the inguinal canal in the spermatic cord and into the abdomen. On the right side, it goes straight into the inferior vena cava, while on the left side, it joins with the renal vein. This can cause problems with draining blood from the left testicle, which is why most varicoceles are found on the left side.[6]

When diagnosing a varicocele, it begins with a clinical examination to assess and determine the stage of the varicocele. Varicoceles are clinically graded on a scale of I to III based on the Dubin and Amlar description. Grade I varicocele is felt only during the Valsalva maneuver, grade II is felt without the maneuver, and grade III is visible from a distance.[7] When we conducted a local examination on our patient, it was observed that while standing, there was a mass in the left scrotum, and the Valsalva maneuver was positive, indicating that this patient has a grade III varicocele according to Dubin and Amlar. Diagnosis relies on clinical observation of dilated and twisted veins in an upright position, with increased prominence during the Valsalva maneuver.[6] A thorough clinical evaluation should additionally involve assessing the size of both testicles to identify any differences in size.[8] For pre-pubertal boys and instances of isolated right varicocele, it is advisable to conduct a renal ultrasound routinely to rule out secondary varicoceles triggered by retroperitoneal tumors extending into the renal vein and inferior vena cava.[9] At present, ultrasound is the most frequently used imaging method for patients with varicocele. Its non-invasive nature, safety for patients, and widespread availability are the primary benefits of this approach.[10] Scrotal ultrasound should be requested to precisely define testicular asymmetry and PRF (Peak Retrograde Flow).[11] PRF is a critical parameter in diagnosis of the varicoceles. Venous reflux into the pampiniform plexus is diagnosed using Doppler ultrasound color flow mapping in both supine and upright positions, along with the Valsalva maneuver. Studies have shown that the severity of reflux observed on Doppler ultrasound correlates with testicular damage.[12]

Sperm analysis, in principle, allows assessment of testicular function, but the World Health Organization (WHO) parameters are not intended for pre-pubertal patients; semen analysis is not widely used, and it is generally recommended only in older adolescents.[13] However, adolescents with varicoceles continue to present a treatment dilemma to the urologist, as objective outcome measures of varicocelectomy (e.g., semen quality or fertility) are not readily attainable or appropriate.[14] In our case, the patient is only nine years old, and not yet showing signs of puberty, and with the consent of the parents, we decided not to perform any semen analysis. Importantly, sperm analysis is of pivotal importance, and it may be requested at least 1.5 years after the onset of puberty.[15]

Microsurgical ligation of varicocele in pre-pubertal children represents a significant advancement in the management of this condition.[16] In adults with infertility, microsurgical subinguinal ligation has become gold standard due to lower of complications and lower post operative recurrences than another technique.[17] By addressing the underlying venous dilation, microsurgical ligation offers a targeted approach to symptom relief while potentially mitigating future complications such as testicular growth impairment and infertility[3]. Although the paternity rate is one of the most important outcomes, it is rarely reported due to the necessity of long-term follow-up. The study by Cayan et al. assessed 286 patients and 122 controls. Patients were treated by microsurgical varicocelectomy.[18] The paternity rate was 77.3% vs. 48.4% (treated vs. untreated), leading the authors to conclude the benefit of treatment in adolescent varicocele. In our patient, we perform microsurgical ligation surgery using optical magnification, which involved a varicocelectomy technique that spared the arteries and lymphatic vessels.

Another surgical management may be considered, including laparoscopic or open surgery.[1,8] The advantages of the subinguinal approach with microsurgical technique are minimal invasiveness with lighter pain, better cosmetic outcomes, and lower recurrence rates. Conversely, the advantages of the retroperitoneal approach include fewer veins to ligate and lower risk if branching of the internal spermatic vein is found at the suprainguinal level.[14] According to the meta-analysis by Silay et al., no difference was found neither between open and laparoscopic techniques nor between subinguinal and high inguinal varicocelectomy. In addition, current evidence does not demonstrate the superiority of any surgical/interventional technique regarding treatment success.[14] Finally, long-term outcomes including paternity and fertility remain unknown and need long-time follow-up.[3,14,19]

After the surgery was performed, there were no more "bags of worms," and during the one-month follow-up, ultrasound Doppler showed no further dilatation of vessels. There was a significant increase in the volume of the left testis from 1.8 cc to 2.3 cc. Subinguinal approaches are often preferred due to their ability to ligate the most distal venous contributions to a clinical varicocele, rather than the use of laparoscopic or retroperitoneal approaches, resulting in a higher relative risk of recurrence and hydrocele formation.[6]

Overall, while microsurgical surgery offers numerous advantages in pediatric patients, including precision, a minimally invasive approach, and reduced risk of recurrence, it also presents challenges related to technical expertise, resource availability, cost, and the learning curve associated with adopting microsurgical techniques. Despite these challenges, the benefits of microsurgical surgery often outweigh the disadvantages, particularly in cases where optimal outcomes and preservation of future fertility are paramount.

CONCLUSION

Microsurgical ligation of varicocele in pre-pubertal children represents a safe and effective therapeutic option, offering symptom relief and potential preservation of future fertility. Further studies are warranted to elucidate the long-term outcomes and reproductive implications of microsurgical varicocelectomy in this population. Nonetheless, the favorable outcome observed in this case supports the use of microsurgical techniques as a first-line treatment approach in symptomatic pre-pubertal children with varicocele.

REFERENCES

1. De Los Reyes T, Locke J, Afshar K. Varicoceles in the pediatric population: Diagnosis, treatment, and outcomes. Can Urol Assoc J. 2017;11(1-2):S34-S39.
2. Riedmiller H, Androulakakis P, Beurton D, Kocvara R, Gerharz E. EAU guidelines on paediatric urology. Eur Urol. 2024;40(5):589-599.
3. Asafu-Adjei D, Judge C, Deibert CM, Li G, Stember D, Stahl PJ. Systematic Review of the Impact of Varicocele Grade on Response to Surgical Management. J Urol. 2020;203(1):48-56.
4. Cannarella R, Calogero AE, Condorelli RA, Giacone F, Aversa A, La Vignera S. Management and treatment of varicocele in children and adolescents: An endocrinologic perspective. J Clin Med. 2019;8(9): 1-11.
5. Siref, Stephen W. Leslie, Hussain Sajjad LE. Varicocele. StatPearls [Internet] Treasure Isl StatPearls Publ. Published online 2024:1-14.
6. Lomboy JR, Coward RM. The Varicocele: Clinical Presentation, Evaluation, and Surgical Management. Semin Intervent Radiol. 2016;33(3):163-169.
7. Bogaert G, van den Heijkant M, Albersen M. Varicocele in Children and Adolescents: A Challenge for Diagnosis and Treatment Indications [Figure presented]. Eur Urol Suppl. 2017;16(8):171-176.
8. Macey MR, Owen RC, Ross SS, Coward RM. Best practice in the diagnosis and treatment of varicocele in children and adolescents. Ther Adv Urol. 2018;10(9):273-282.
9. Gleason A, Bishop K, Xi Y, Fetzer DT. Isolated right-sided varicocele: Is further workup necessary? Am J Roentgenol. 2019;212(4):802-807.
10. Lorenc T, Krupniewski L, Palczewski P, Go??biowski M. Warto?? ultrasonografii w diagnostyce ?ylaków powrózka nasiennego. J Ultrason. 2016;16(67): 359-370.
11. Poon SA, Gjertson CK, Mercado MA, Raimondi PM, Kozakowski KA, Glassberg KI. Testicular Asymmetry and Adolescent Varicoceles Managed Expectantly. J Urol. 2010;183(2):731-734.
12. Vanlangenhove P, Dhondt E, Everaert K, Defreyne L. Pathophysiology, diagnosis and treatment of varicoceles: A review. Minerva Urol e Nefrol. 2014;66(4):257-282.
13. Ku JH, Kim SW, Park K, Paick JS. Benefits of microsurgical repair of adolescent varicocele: Comparison of semen parameters in fertile and infertile adults with varicocele. Urology. 2005;65(3):554-558.
14. Silay MS, Hoen L, Quadackaers J, et al. Treatment of Varicocele in Children and Adolescents: A Systematic

Review and Meta-analysis from the European Association of Urology/European Society for Paediatric Urology Guidelines Panel(Figure presented.). Eur Urol. 2019;75(3):448-461.
15. Apriansah IM. Testicular Volume to Semen Analysis in Male Patients with Andrology Poly, RSUD Dr. Soetomo Surabaya. 2022;3(2):34-37.
16. Schiff J, Kelly C, Goldstein M, Schelgel P, Poppas D. Managing varicoceles in children: Results with microsurgical varicocelectomy. BJU Int. 2005;95(3):399-402.
17. Parrilli A, Roberti A, Escolino M, Esposito C. Surgical approaches for varicocele in pediatric patient. Transl Pediatr. 2016;5(4):227-232.
18. Çayan S, ?ahin S, Akbay E. Paternity Rates and Time to Conception in Adolescents with Varicocele Undergoing Microsurgical Varicocele Repair vs Observation Only: A Single Institution Experience with 408 Patients. J Urol. 2017;198(1):195-201.
19. Piotr Dobronski, Karolina Dobronska, Lukasz Kupis PR. Subinguinal microsurgical varicocelectomy is safe and effective in a solitary testicle. Asian J Androl. 2019;21(July):1-4.