Original
Original

Endoscopic Balloon Dilatation for Treatment of Primary Obstructive Megaureter: Experience of a Center

Dilatação Endoscópica de Balão para Tratamento de Megaureter Obstrutivo Primário: Experiência de um Centro

Sofia Morão[1], Vanda Pratas Vital[1], Dinorah Cardoso[1], Fátima Alves[1], Filipe Catela Mota[1], João Pascoal[1]

Abstract

Introduction: *Congenital obstructive megaureter may be treated with endoscopic balloon dilatation, particularly in children under one year of age. We report our experience over a six year period.*

Methods: *All patients with diagnosis of primary obstructive megaureter treated with endoscopic balloon dilatation from 2009 to 2014 (6 years) were included. The diagnosis of primary obstructive megaureter was based on dilatation of the distal ureter greater than 7 mm, obstructive curve on MAG-3 diuretic renogram and absence of vesicoureteral reflux. After diagnosis, conservative management was maintained with antibiotic prophylaxis in all patients. The indications for surgery were a combination of clinical, ultrasonographic and renographic findings. Under general anesthesia and after retrograde ureteropielography, high pressure balloon dilation of the ureterovesical junction was performed under direct and fluoroscopic vision until the disappearance of the narrowed ring. A double-J catheter was positioned. Follow-up was performed with ultrasonography and diuretic renogram. The success of the intervention was defined by improvement of hydroureteronephrosis (at least 2 grades).*

Results: *A total of nine patients underwent this procedure on a single ureter, two girls and seven boys, with a mean age of 7.6 months (range 1-14) at the intervention. Five were left sided and four were right sided. All patients had prenatal diagnosis of hydroureteronephrosis. No patients were lost to follow-up (average 46.7 months). They all had hydroureteronephrosis greater than grade 3 and preoperative MAG-3 diuretic renogram was obstructive in all cases. Mean differential function of the affected kidney was 46.2% (range 40-53%). The main indication for surgical treatment was progressive hydroureteronephrosis. All patients were treated endoscopically with no intraoperative complications. Ultrasound showed improvement of the hydroureteronephrosis in six patients (66.7%). Three patients were reimplanted (33.3%). The mean differential renal function after the procedure was 47.4% (range 41-53%). At the latest follow-up assessment, all patients remained asymptomatic.*

Resumo

Introdução: O megauretero obstrutivo congénito pode ser tratado por dilatação endoscópica com balão, especialmente nas crianças com menos de um ano de idade. Este trabalho revela a nossa experiência num período de seis anos.

Métodos: Todos os doentes com o diagnóstico de megauretero obstrutivo congénito submetidos a dilatação endoscópica com balão foram incluídos, no período de tempo compreendido entre 2009 e 2014 (seis anos). O diagnóstico baseou-se na identificação de dilatação do uretero distal superior a 7 mm, padrão obstrutivo no renograma MAG-3 e ausência de refluxo vesico-ureteral. Após o diagnóstico, adoptou-se uma atitude conservadora com início de profilaxia antibiótica e vigilância ecográfica em todos os doentes. A indicação cirúrgica resultou de um conjunto de achados clínicos, ecográficos e renográficos. Sob anestesia geral, e após pielografia retrógrada, foi realizada dilatação endoscópica da junção uretero-vesical sob visão directa e controlo fluoroscópico até ao desaparecimento do anel estenótico, colocando-se no final um *stent* duplo J. O seguimento foi feito com controlo ecográfico e renograma. O sucesso da intervenção cirúrgica foi definido como uma melhoria no grau de hidroureteronefrose (pelo menos 2 graus).

Resultados: No total, 9 doentes foram intervencionados num só ureter, duas meninas e sete meninos, com idade média de 7,6 meses (entre 1-14) na data da cirurgia. Cinco foram no lado esquerdo e quatro no lado direito. Todos os doentes tinham diagnóstico pré-natal de hidroureteronefrose. Nenhum doente foi perdido no seguimento (média 46,7 meses). Todos tinham hidroureteronefrose de grau superior ou igual a 3 e padrão obstrutivo no renograma MAG-3. A função relativa média do rim homolateral foi 46,2% (entre 40-53%). A principal indicação cirúrgica foi o agravamento da hidroureteronefrose durante a vigilância ecográfica. Não existiram complicações intra-operatórias. A ecografia mostrou melhoria da hidroureteronefrose em seis doentes (66,7%). Três doentes foram re-implantados (33,3%) A função relativa média após a cirurgia foi 47,4% (entre 41-53%). Na consulta de seguimento mais recente, todos os doentes estavam assintomáticos.

[1]Pediatric Urology Unit, Department of Pediatric Surgery, Hospital Dona Estefânia, Centro Hospitalar Lisboa Central, Lisbon, Portugal

Conclusion: Endoscopic balloon dilatation is a useful option in the management of primary obstructive megaureter requiring surgical intervention and may be considered first line treatment in small children.

Keywords: Child; Constriction, Pathologic; Dilatation; Endoscopy; Hydronephrosis; Ureter/abnormalities; Ureteral Obstruction.

Conclusão: A dilatação endoscópica com balão é uma opção na abordagem do megauretero obstrutivo primário com indicação operatória e pode ser considerado como tratamento de primeira linha nas crianças com menos de um ano de idade.

Palavras-chave: Criança; Constrição Patológica; Dilatação; Endoscopia; Hidronefrose; Obstrução Ureteral; Ureter/anomalias congénitas.

Introduction

Megaureter is not a diagnosis but a descriptive term for a dilated ureter.[1] A retrovesical ureteric diameter greater than 7 mm from 30 week's gestation onwards is abnormal and should be investigated postnatally.[2]

Smith classified megaureters into four categories: obstructed, refluxing, refluxing with obstruction and non-refluxing/non-obstructed. These were later subdivided by King into primary and secondary.[2]

Primary obstructive megaureter (POM) is most commonly caused by a distal adynamic ureteral segment. A few histopathologic characteristics of the adynamic segment have been described such as abnormal neuromodulation, abnormal collagen deposition and abnormalities in the interstitial cells of Cajal.[3] The timing of smooth muscle differentiation in the distal ureter is unknown but may be the key to explain pathogenesis of primary (or congenital) megaureter.

The process whereby the circular muscle pattern, which is typical of the fetal ureter, changes progressively into double muscle layers on the full term infant, may last up to two years,[2] and may explain why this condition resolves spontaneously in approximately 80% of those patients diagnosed prenatally and that is why conservative management is safe initially.[4]

Surgical management is indicated when initial differential renal function (DRF) is less than 40% and when conservative management fails.[2] Traditionally, the surgical management of POM has been via ureteral reimplantation with or without ureteral remodeling. However, in newborns and children under one year of age this technique is very difficult and has a high complication rate. Temporary cutaneous diversion may be beneficial in this cases but it will require two or more procedures for correction.

With advent of minimally invasive surgery, in 1998, Angulo *et al* published the first report of endoscopic balloon dilatation for POM in children and, since then, several publications have shown that the procedure is feasible, safe and less invasive in very young patients.[5]

This study assesses the long-term effectiveness of endoscopic balloon dilatation in a case-series of six years, in order to evaluate if it can be considered as an alternative to ureteral reimplantation in small children with POM.

Methods

All patients with diagnosis of POM treated with endoscopic balloon dilatation from 2009 to 2014 were included. Data from clinical records was retrieved.

The diagnosis of POM was based on the following parameters: dilatation of the distal ureter greater than 7 mm, obstructive curve on MAG-3 diuretic renogram and absence of vesicoureteral reflux (VUR). Ultrasound was employed to measure the diameter of renal pelvis, distal ureter and the characteristics of renal parenchyma. The degree of HUN was defined in accordance with guidelines of the Society for Fetal Urology.[6-8] Voiding cystourethrogram (VCUG) was performed to rule out VUR. On MAG-3 diuretic renogram, obstruction was defined as persistence of more than 50% of the tracer in the renal pelvis 20 minutes after diuretic administration.

After the initial diagnosis, conservative management was maintained with antibiotic prophylaxis (oral trimethoprim 1% 0.1 mL/kg, once a day) in all patients. The indications for surgery were a combination of clinical, ultrasonographic and renographic findings with at least one of the following conditions: initial DRF < 40% with an obstructive excretion pattern on renogram MAG-3, progressive HUN or febrile urinary tract infection (UTI) (Table 1).

Under general anesthesia, cystoscopy was performed with a Wolf® 9Fr pediatric cystoscope (Knittlingen, Germany) with a 5Fr working channel. The ureterovesical junction was passed with a flexible guidewire (0.014 mm Terumo®) introduced up to the renal pelvis which was then filled with radiologic contrast (Fig. 1A), confirming the diagnosis of megaureter and the defining anatomy. A Biosensors Powerline® dilating balloon catheter (4 mm diameter and 30 mm length) was insufflated to 12 or 14 atmospheres until the stenotic ring was no longer present (with 3 to 6 minutes of waiting time). The stenotic ring was always visible before dilatation. Then, distal ureterography was performed, confirming dilatation of the stenotic segment and a 4-4.8Fr 10-14 cm double-J stent was left in place (Fig. 1B). The bladder catheter was withdrawn after 24 hours of surgery and the patient was discharged after 24-48 hours, depending on haematuria. The double-J stent was removed under general anesthesia after a mean period of 5.4 months

Table 1: Patients characteristics and results

Patient number	Age at surgery (months)	Diagnosis	HUN grade	% DRF	Indications for surgery	Postop HUN grade	Postop % DRF	Ureteral reimplantation	Follow-up (months)
1	1	prenatal	3	–	increasing HUN	0	49	no	72
2	14	prenatal	3	49	increasing HUN	0	49	no	60
3	13	prenatal	3	40	DRF < 40	3	45	yes - recurrent febrile UTI	60
4	2	prenatal	4	44	increasing HUN	1	44	no	60
5	9	prenatal	3	–	increasing HUN	0	49	no	48
6	1	prenatal	3	–	increasing HUN	3	–	yes - persistent HUN + febrile UTI	48
7	7	prenatal	4	45	increasing HUN	4	49	yes - increasing HUN	48
8	12	prenatal	3	–	increasing HUN	0	41	no	12
9	9	prenatal	4	53	increasing HUN	2	53	no	12

(range: 2-7 months).

Follow-up included renal ultrasound at one, three, nine and twelve months and annually thereafter; MAG-3 diuretic renogram at six months, two and five years. VCUG was not performed routinely. Antibiotic prophylaxis was maintained until stent was removed.

The success of the intervention was defined by improvement of HUN (at least 2 grades). Success rate was calculated with 95% confidence interval (95% CI) using openEPI.[9,10] Data on upper urinary tract infections and further surgical interventions were collected.

Results

Out of 110 cases of diagnosed megaureters on that period, a total of nine patients underwent this procedure on a single ureter, two girls and seven boys, with a mean age of 7.6 months (range 1-14) at the intervention. Five were left sided and four were right sided. All patients had prenatal diagnosis of HUN. No patients were lost to follow-up (average 46.7 months; standard deviation 21.2) (Table 1).

They all had HUN greater than grade 3. VCUG showed existence of Hutch's diverticulum in two patients and none had VUR. Preoperative diuretic renogram was obstructive in all patients in whom it was performed (patients 2, 3, 4, 7 and 9). Mean differential function of the affected kidney was 46.2% (range 40-53%).

The indication for surgical treatment was progressive HUN in eight cases and deterioration of renal function (DRF less than 40%) in one case (Table 1).

All nine patients were treated endoscopically with no intraoperative complications. There was one case of stent migration to the distal ureter that was removed endoscopically.

Ultrasound showed improvement of the HUN in six patients (Fig. 2). The success rate was 66.67% (95% CI) (Table 1).

Three patients were reimplanted (one Cohen and two Politano-Leadbetter neoureterocistostomy). Patients 3 and 6 were reimplanted because of recurrent urinary infections, two and six months after endoscopic dilatation, respectively. One patient was reimplanted twenty-one months after endoscopic balloon dilatation because of worsening of HUN with persistence of the obstructed pattern in a diuretic renogram.

The mean DRF after the procedure was 47.4% (range 41-53%). The mean follow-up was 46.7 months (range 12-72) (Table 1).

At the most recent follow-up assessment, all patients remained asymptomatic. The two patients with shorter follow-up were assessed at about two years of age. Both had great improvement of HUN without obstructive pattern at the postoperative MAG-3 renogram.

Discussion

In this case-series, the primary success rate was 66.7%. In three cases (33.3%) the ureter was reimplanted. The secondary success rate (after reimplantation) was 100%. These results are similar to those previously published.

In 1998 Angulo *et al* and in 2007 Angerri *et al* described the first POM treatment with endoscopic balloon dilatation, which

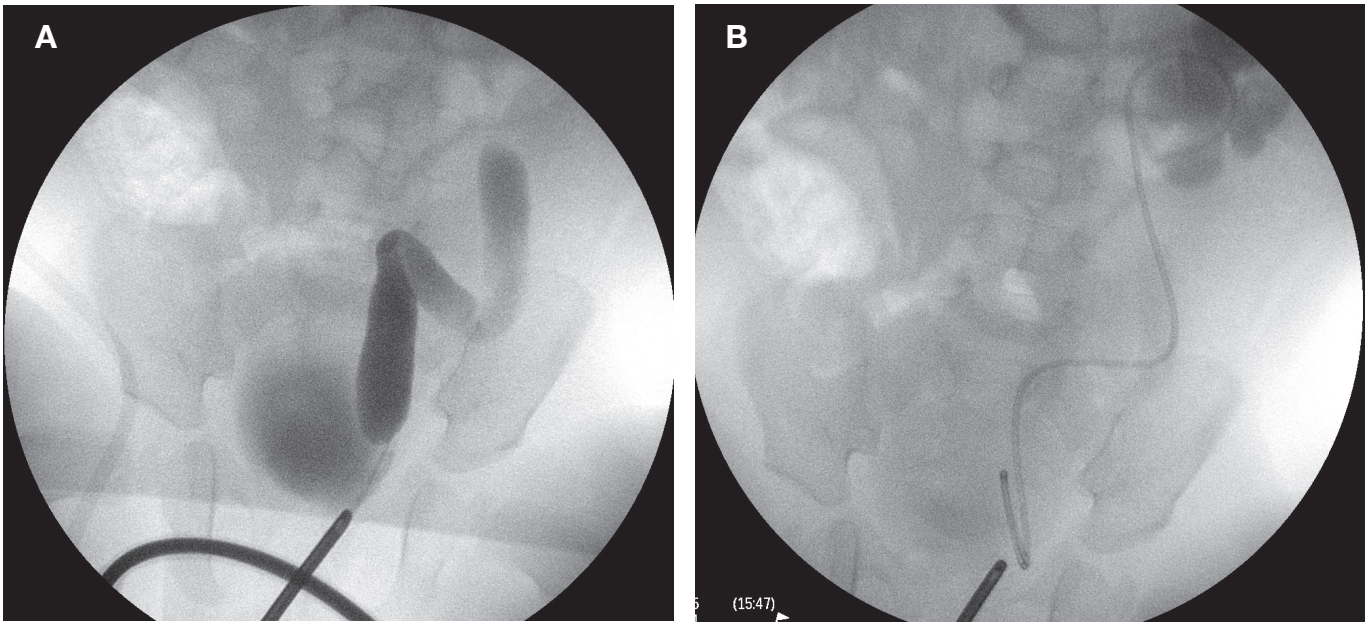

Figure 1: Cystoscopy. A: Contrast injection B: stent positioning.

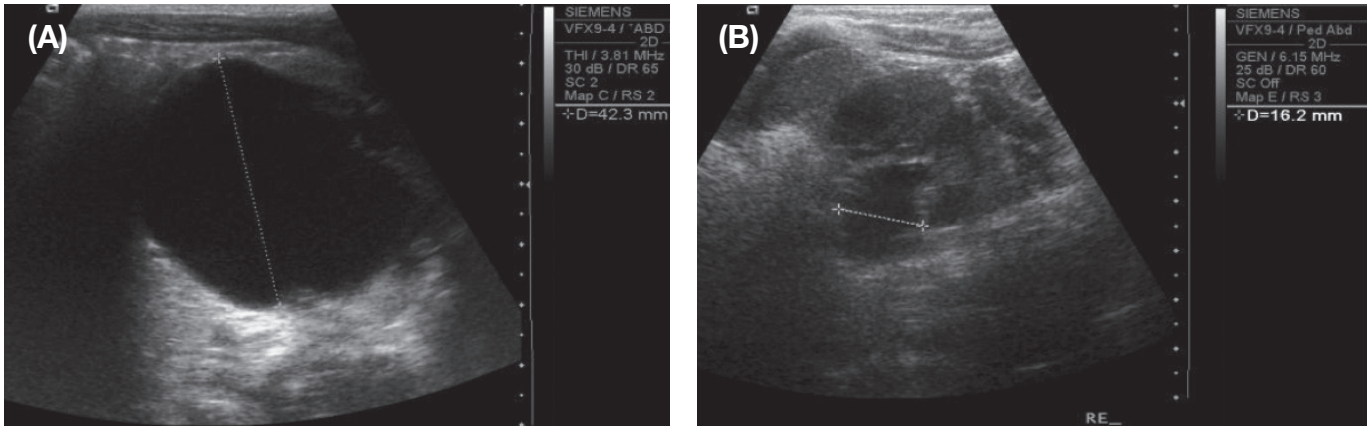

Figure 2: Renal pelvis at ultrasound before (A) and after (B) endoscopic balloon dilatation.

was believed to be a definitive, safe and less invasive treatment that avoids unnecessary open surgery.[5] A small number of publications have reported short series with good results in the short and medium-term. García-Aparicio *et al*[11] presented a series of thirteen patients with postoperative improvement of HUN in 84.6% (eleven cases) and need for reimplantation in 23% (three cases). Carroll *et al*[3] presented a series of thirty-one patients with improvement of HUN in 48.4% (fifteen cases) and reimplantation in 35.4% (eleven cases).

Historically, the surgical management of POM has been based on ureteral reimplantation with or without ureteric remodeling, with a reported success rate of 90-96%.[5] However, ureteric reimplantation in infants less than one-year of age may be challenging due to the discrepancy between the grossly dilated ureter and the small infantile bladder and the concern regarding possible iatrogenic bladder dysfunction, such as VUR.[2]

Therefore, some authors consider alternative temporizing or permanent interventions avoiding reimplantation in infancy.

In the era of minimally invasive surgery, endoscopic management of POM offers a less invasive option for treatment. Furthermore, should an endoscopic approach fail, reimplantation surgery can still be performed.[10]

The youngest patient in the series of Angerri *et al* was twelve months, in Garcia-Aparicio *et al* was four months, in Carroll *et al* was two months and in Bujon *et al* was one month. In the present series the youngest patient subject to intervention was one month old.[3-5,11]

The stents were left for more time than the average mentioned on literature (2 months) because of the need of general anesthesia and lack of operating room availability.

Virtually no morbidity was detected, with only one case of stent migration without clinical implications.

This study has a small number of cases and short follow--up time which limits conclusions, however the results show that endoscopic balloon dilatation is a useful option in the management of POM requiring surgical intervention, reducing morbidity associated with open surgery without sacrificing efficacy.

Conclusion

This study shows that endoscopic balloon dilatation of POM is a safe, feasible and less invasive procedure with few post-operative complications and may be considered first line treatment in the management of POM in small children.

Ethical Disclosures
Conflicts of interest: The authors report no conflict of interest.

Funding sources: No subsidies or grants contributed to this work.

Protection of Human and Animal Subjects: The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of Data: The authors declare that they have followed the protocols of their work center on the publication of patient data.

Responsabilidades éticas
Conflitos de Interesse: Os autores declaram a inexistência de conflitos de interesse na realização do presente trabalho.

Fontes de Financiamento: Não existiram fontes externas de financiamento para a realização deste artigo.

Protecção de Pessoas e Animais: Os autores declaram que os procedimentos seguidos estavam de acordo com a Declaração de Helsínquia da Associação Médica Mundial.

Confidencialidade dos Dados: Os autores declaram ter seguido os protocolos do seu centro de trabalho acerca da publicação dos dados de doentes.

Corresponding Author / Autor Correspondente
Sofia Morão - sophia.morao@gmail.com
Department of Pediatric Surgery, Hospital Dona Estefânia, R. Jacinta Marto, 1169-045 Lisboa, Portugal

Recebido/Received: 2017-05-08
Aceite/Accepted: 2017-06-20

REFERENCES

1. Christensen T, Coplen D. Ureteral obstruction and malformations In: Holcomb G, Murphy J, editors. Ashcraft's Pediatric Surgery. 5th ed. Philadelphia: Saunders Elsevier; 2010. Chapter 55
2. Farrugia MK, Hitchcock R, Radford A, Burki T, Robb A, Murphy F, et al. British Association of Paediatric Urologists consensus statement on the management of the primary obstructive megaureter. J Pediatr Urol. 2014; 10: 26-33.
3. Carrol D, Chandran H, Joshi A, McCarthy, Parashar K. Endoscopic placement of a double-J ureteric stents in children as a treatment for primary obstructive megaureter. Urol Ann. 2010; 2: 114-8.
4. García-Aparicio L, Blásquez-Gómez E, Martin O, Palazón P, Manzanares A, García-Smith N, et al. Use of high-pressure balloon dilatation of the ureterovesical junction instead of ureteral reimplantation to treat primary obstructive megaureter: is it justified? J Pediatr Urol. 2013; 9: 1229-33.
5. Bujons A, Saldaña L, Caffaratti J, Garat JM, Angerri O, Villavicencio H. Can endoscopic balloon dilatation for primary obstructive megaureter be effective in a long term follow-up? J Pediatr Urol. 2015; 11: 36e1-37.e6.
6. Nguyen H, Benson C, Bromley B, Campbell J, Chow J, Coleman B. Multidisciplinary consensus on the classification of prenatal and postnatal urinary tract dilation (UTD classification system). J Pediatr Urol. 2014; 10: 982-99.
7. Timberlake D, Herndon D. Mild to moderate postnatal hydronephrosis--grading systems and management. Nat Rev Urol. 2013; 10: 649-56.
8. Kennedy W. Assessment and management of fetal hydronephrosis. Urol Neorev. 2002; 3:10.
9. Dean AG, Sullivan KM, Soe MM. OpenEpi: Open Source Epidemiologic Statistics for Public Health, Version3. [assessed on 2015/11/19] Available from: http://www.OpenEpi.com
10. Christman M, Kasturi S, Lambert R, Kovell R. Endoscopic management and the role of double stenting for primary obstructive megaureters. J Urol. 2012; 187: 1018-23.
11. García-Aparicio L, Rodo J, Krauel L, Palazon P, Martin O, Ribó JM. High pressure balloon dilatation of the ureterovesical junction – first line approach to treat primary obstructive megaureter? J Urol. 2012; 187: 1834-8.