

Uretero-pelvic junction obstruction in children: Is vascular hitch an effective and safe solutions in very long term outcome? Report of 25 years follow-up

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Abstract

Vascular (VH) according to Hellstrom-Chapman technique is considered a safe and effective alternative approach to pure extrin-

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sic Ureteropelvic Junction Obstruction (UPJO) with good results in short and medium term, but few data are available on long and very-long term outcomes. Our aim is to evaluate VH long and very-long term outcomes in patients treated in pediatric age focusing on relapse, development of hypertension and/or inferior polar kidney hypotrophy during puberty and adulthood. From 1990 to 2015 in our Department 76 children were treated by open or laparoscopic VH for pure extrinsic-UPJO. We were able to contact 54 of 76. 41 patients (25 males, 16 females) accepted to be studied. Mean follow-up time was 12.7 years (range 6-27 years); mean age at the assessment was 22.2 years. We excluded patients who were younger than 13 (if females) or 14 (if males) at the assessment (upper limits of physiological puberty onset). Patients were followed with US, MAG-3-scan and arterial blood pressure measurement. Collected data were compared with the preoperative ones by Student *t*-test. 95% of US images and MAG-3-scan reports were compatible with complete resolution of obstruction with good renal functionality. 87% of patients were completely healthy. We recorded 3 cases of hypertension (7%) not secondary to renovascular origin; 2 cases with recurrent flank pain (5%) with slightly dilated pelvis at the US and sub-obstructive pattern at MAG-3-scan with preserved renal function. Our experience confirms that VH, (open/laparoscopic) is a safe and effective procedure with good outcomes at very long-term follow-up. No patients at puberty and in adulthood required reoperation or presented polar hypotrophy and related vascular hypertension. VH is an alternative approach to pure extrinsic-UPJO. There were few data about long and very-long term outcomes in patients after this kind of surgery. We followed-up 41 patients confirming that VH (open/laparoscopic) is safe and effective with good long-term outcomes.

Introduction

Uretero-Pelvic Junction Obstruction (UPJO) may be caused by intrinsic ureteral stenosis (aperistaltic dysplastic segment of UPJ) or by extrinsic compression from Crossing Vessels (CV). UPJO due to CV is a rare condition in neonates and has the highest incidence in older children, presenting the typical "polar vessels syndrome" including absent/irrelevant renal pelvis dilatation at prenatal ultrasound and a late presentation with intermittent signs (vomiting, flank pain or renal colic). Dismembered pyeloplasty according to Anderson and Hynes (AHDP)¹ is the gold standard treatment for most intrinsic and extrinsic UPJO. In 1949, Hellström *et al.*² described an alternative approach to pure extrinsic-UPJO: it involved displacing the lower pole vessels cranially and then anchoring them to the anterior pelvic wall using vascular adventitial sutures. Chapman³ modified this tech-

nique by securing a more superior position of the lower pole vessels within a wrap of the anterior redundant pelvic wall without vascular adventitial sutures. This technique has since been used in children as an alternative to AHDP with good results in the short and medium term outcomes^{4,5} when criteria described in the table are respected (Table 1).⁶⁻⁸ Literature about long and very-long term outcomes is poor⁹ and some authors claim that inferior polar vessels can narrow after the surgical transposition due to the patient's growth.^{10,12} According to this theory, Vascular Hitch could be related with development of hypertension at puberty and adulthood and/or with inferior polar hypotrophy due to prolonged blood hypoperfusion.

Materials and Methods

338 patients were treated for UPJO in our Department from 1990 to 2015. 76 of these children were treated successfully by open or laparoscopic VH for pure extrinsic-UPJO by crossing vessels. The technique was the same for all patients and consisted in the exposure of the UPJ and Crossing Vessels (CV), their dissection and mobilization followed by diuretic test administering a bolus of normal saline (20 mL/kg IV) before the complete CV mobilization. Furosemide (1 mg/kg IV) was administered after completed mobilization. The UPJ was inspected to exclude any intrinsic visible stenosis and visualizing the peristalsis associated with an easy urine passage across the junction after temporarily CV transposition. Completed the test successfully vascular hitch was performed. In all patients the UPJ-ureter anatomical relation corresponded to Schneider's third type.¹³ The intraoperative criteria to perform VH were: i) presence of lower pole CV; ii) normal UPJ on inspection; iii) response to DT with emptying of the dilated pelvis after vessel displacement.^{5,7} We were able to contact 54 of these 76 and 41 patients (25 males, 16 females) accepted a complete follow up. Our assessment focused on: i) clinical examination

(presence/frequency of episode of flank or abdominal pain); ii) renal US with Doppler (persistence of sonographic improvement of hydronephrosis, homogeneous kidney vascularization); iii) MAG-3 scan (no impairment of drainage curves on renograms, stable/improved renal function); iv) measurement of arterial Blood Pressure (BP). The goal of study was absence of symptoms, normal BP/no development of renovascular hypertension, absence of kidney lower pole hypotrophy. We reviewed the charts of patients to determine mean age at surgery, preoperative imaging results, operative time, need for conversion, hospital stay length, intraoperative and postoperative complications. We excluded in this study a patient who needed a corrective AHDP two years after VH and all patients younger than 13 (if females) or 14 (if males) at the study: upper limits of puberty physiological onset. This to avoid possible bias due pre-pubertal stage. Patient's characteristics are reported in the Table 2.

Blood pressure measurement and renal US with Doppler were performed in our Department. MAG-3-scans were performed at Nuclear Medicine Department of our Hospital. The antero-posterior or pelvic diameter(mm) at US evaluation and the ipsilateral renal functionality (%) at MAG-3-scan were compared with the preoperative data by Student *t*-test. A p-value <0.05 was considered statically significant.

Results

95% of US images and MAG-3 scan reports were compatible with complete resolution of UPJO and good renal function. Mean antero-posterior pelvic diameter was 11mm at the study point, significantly reduced if compared to the mean preoperative value of 33mm (p value <0.001). Parenchymal echogenicity was altered only in one case, a 15-years-old boy operated at 9 years of age. His AP pelvic diameter was 40mm before surgery and 7mm at the study point. His ipsilateral renal function at MAG-3 scan was 24%

Table 1. Criteria to perform a safe vascular hitch.

Five criteria for performing a safe vascular hitch

1. Typical polar vessels syndrome, including absent/irrelevant renal pelvis dilatation at prenatal ultrasound and a late presentation with intermittent signs (vomiting, flank pain or renal colic)
2. Marked hydronephrosis at the US when the pain is present or a positive Water load and Diuretic Ultrasound Test (WDUT) as standardized in our department
3. Obstructive pattern on MAG-3 scan
4. Positive intraoperative Diuretic Test (DT) as standardized in our institute since 1999⁹
5. Magnetic Resonance Urography (MRU) is usually done in our institute to confirm the presence of polar vessels¹⁰⁻¹²

Table 2. Patient's characteristics.

Patients characteristics (n. 41)

Mean age at surgery	9.5 years (SD 4.08)
Mean follow-up time	12.7 years (SD 6.68 months)
Mean age at assessment	22.2 years (SD 7.28)
Males/females	25/16
Open/laparoscopic approach	15/26
Need of conversion	0/26
Right side/left side	14/27
Mean operative time for open/laparoscopic procedure	70/120 minutes
Mean hospital stay length for open/laparoscopic procedure	6/2.5 days

before surgery and 23% at the study point, with no evidence of obstructive pattern (Figure 1a).

Mean ipsilateral renal function at MAG-3 scan was 46% at the study point, compared to a mean value of 40% before surgery (p value <0.001). There was no evidence of hypoperfusion of inferior renal pole (Figure 1b).

Two patients report recurrent flank pain on the operated side, after a long period of postoperative clinical well-being. The first patient is a 17-year-old girl with a history of prenatal diagnosis operated by laparoscopic approach when she was 7. The symptoms arose 9 years after surgery. Ipsilateral renal function is 36% at MAG-3 scan, similar with that described in the preoperative scan (36%) and with the postoperative one performed two years after the operation (38%). MAG-3 scan report described obstructive pattern. Antero-posterior pelvic diameter was 33mm at US. A retrograde pyelography showed the presence of pyelic and calyceal dilation and a short narrowing at the pyeloureteral junction. Dilatation was carried out by a balloon catheter (8mm, 16 Atm) with resolution of the stenosis confirmed by the "balloon test". A double-J ureteral stent was placed and removed after a month. At one-year follow-up the patient is clinically well.

The second patient is a 27-year-old girl who was operated by 'open' approach at 12-years of age. Symptoms arose 12 years after the operation and reported occasionally contextually to the first days of menstruation. Dynamic renal scintigraphy reveals 45% differential renal function and partial obstructive pattern. Antero-posterior pelvic diameter was 28mm at US. He denied consent to endoscopic evaluation for diagnostic/therapeutic approach.

In three patients was found arterial hypertension that was carefully investigated to evaluate renovascular origin related to surgery: one 23-years-old man operated at 3years of age by 'open' approach, one 16-years-old boy operated at 6 years of age and a 15-year-old boy operated at 7years of age by laparoscopic approach. The diagnosis of hypertension was confirmed by 24h blood pressure monitoring, which revealed systolic/diastolic blood pressure values higher than 95th percentile. The patients underwent nephrological and cardiological examinations. They were studied with blood examination including blood count, ionemia, renal function, thyroid function, ACTH-cortisol axis, renin, aldosterone, urinary catecholamines, urinalysis; ECG; US-Color-Doppler of renal arteries and *fundus oculi*.

The 23-years-old patient (BMI 25.1) reports frequent

headaches that respond to NSAID therapy and episodes of flushing and palpitations. It reports important family cardiological history with an episode of myocardial infarction of the paternal uncle at 20-years-old and the paternal grandfather at 50. US was normal, MAG-3 scan was normal (differential renal function 49%). CBC, ionemia, renal function, thyroid function, ACTH cortisol axis, renin, aldosterone, urinary catecholamines, urinalysis: normal. After failure of therapy with ACE inhibitor (Ramipril 10 mg) he is currently on therapy with Ca²⁺ antagonist (Amlodipine 10 mg).

The Doppler of the 15-years-old patient (BMI 30.7) found an accelerated peak flow (100 cm/s) at the distal portion of renal artery on the operated side, so the patient underwent renal MRI angiography that no highlights endoluminal signal defects in the main renal arteries or in the transposed polar artery. MAG-3 scan does not demonstrates uptake defects in the perfusion phase on the operated side. Ipsilateral renal functionality on the operated side was 47% with no obstructive pattern. Parenchymal echogenicity was not altered. Pelvic AP diameter was 12 mm. CBC, ionemia, renal function, thyroid function, ACTH-cortisol axis, renin, aldosterone, urinary catecholamines, and urinalysis: normal. The patient is currently in therapy with Ca²⁺ antagonists (Amlodipine 5 mg).

The 16-years-old patient (BMI 31.6) presents antero-posterior pelvic diameter of 19mm at US, non-obstructive pattern and 42% differential renal function on the operated side on MAG-3 scan. CBC, ionemia, renal function, thyroid function, ACTH-cortisol axis, renin, aldosterone, catecholamines urinary tract, urinalysis: normal. He is currently in therapy with Ca²⁺ antagonist (Amlodipine 5 mg).

Follow-up data are reported in Table 3.

Discussion

Literature about long-term follow-up after open or laparoscopic VH is currently poor. A 2015 study¹ demonstrates 96% of effectiveness of the procedure on a series of 70 patients with mean follow-up time of 52 months. They evaluated decreasing in pyelic dilation (from preoperative mean value of 33mm to 9mm at study-point, p value <0.001) and absence of obstructive pattern on dynamic renal scintigraphy (absent at study point in 53/56 patients). This study did not investigate the correlation between

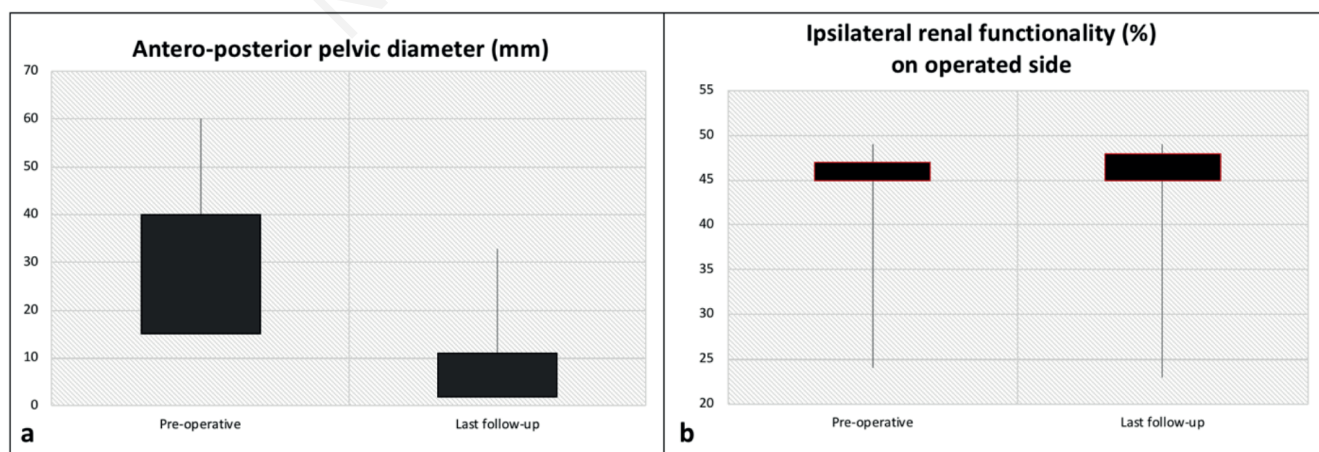


Figure 1. a) Antero-posterior pelvic diameter. Comparison between the preoperative value (left) vs. value at the study point, significantly reduced (right); b) Mean ipsilateral renal function at MAG-3 scan. Comparison pre-operative mean value (46%) and the study point value (40%) (p value <0.001).

VH and development of hypertension. The same group published a study in 2016 with mean follow-up time of 67.2 months based on 33 patients. No cases of hypertension have been reported except in one patient with pre-operative diagnosis, treated by ACE inhibitors e Ca²⁺-antagonists. Renal perfusion was investigated in 17 patients with color-Doppler that did not show differences in the vascularization of the lower pole compared to the remaining parenchyma. The same study reported 3 cases of recurrent flank pain on the operated side. Follow-up interval of these patients was 98, 81 e 98 months. Only one of these had a dilated pelvis (AP diameter 31mm). The authors excluded that in the other two cases the symptoms could recognize renal cause.

Our mean follow-up time (152.4 months, more than 12 years) is currently the longest among those reported in literature (Table 4).

Renal US confirmed the absence of relapse and the very-long term effectiveness of the procedure. The medium antero-posterior diameter of the pelvis decreased from 33 mm (preoperative value) to 11mm at study point (p value <0.001).

By a revision of Literature three retrospective studies with short follow-up analyzed Doppler renal ultrasound postoperatively without evidence of lower pole perfusion defects although statural growth.¹³⁻¹⁶ The use of MAG-3 scan confirmed UPJO resolution and evaluated the perfusion phase of the lower pole of the operated kidney with greater sensitivity than color-Doppler. In the present study, all patients who underwent parenchyma perfusion analysis with Doppler renal ultrasound showed homogeneous vascularization of the kidney.

We did not demonstrate any uptake defects except in one only case (1/41), in a totally non-functioning kidney. This finding may excluded the possibility that the vascularization of the lower pole may suffer during and after growth due to a possible strain and compression of the transposed vessel, even in the absence of symptoms. The research for a correlation between the Hellstrom-Chapman technique and the development of hypertension in adult-

hood led us to carefully investigate the blood pressure of enrolled patients. The hypothesis underlying the suspicion is that during puberty the growth modifies the anatomy of the transposition, stretching the abnormal polar vessel and overstimulating the renin-angiotensin-aldosterone axis. Furthermore we must consider that according to pediatric nephrologists hypertension with PUJ obstruction predominantly occurs in the second decade of life.^{17,18}

The three cases in which Holter-24h measurement confirmed the presence of arterial hypertension were referred for internal consultancy. The laboratory and imaging findings excluded a renovascular cause of disease. Diagnosis of essential hypertension was made. These findings agreed with the other data currently present in Literature, suggesting that even in very long term follow-up there is no correlation between Hellstrom-Chapman technique and development of hypertension in puberty and post-puberty age.

The case of recurrent flank pain that arose 9 years after surgery was carefully investigated, also in invasive way, to excluded two hypotheses: i) the transposition failed and polar vessels moved down obstructing the ureter again; ii) there was an intrinsic stenosis not recognized during surgery. The story of clinical well-being for many years after surgery, the pyelographic appearance, the obstructive pattern on scintigraphy without loss of kidney function suggested that pain origin was a recent short fibrotic stenosis maybe caused from minimal vesicoureteral reflux. She underwent also to cystourethrography, which revealed a minimal vesicoureteral reflux. During the pyelography, our surgeons performed a pneumatic dilatation and the symptoms have now completely regressed.

Finally, we did not found statistically significant differences between open and laparoscopic approach in very-long term outcomes. Therefore, with the same efficacy and safety, our gold standard is the laparoscopic approach for less invasiveness, better aesthetic outcome, less need for days of hospital stay after surgery (6 days for open approach vs. 2.5 for laparoscopic one), less need for

Table 3. Follow-up data.

Follow-up results	
Mean follow-up time (minimum-maximum)	152.4 months (72-324)
Mean age at follow-up (minimum-maximum)	22.2 years (13-37)
Clinical findings	
Recurrent flank pain	2/41 (5%)
Arterial hypertension	3/41 (7%)
US findings	
Altered parenchymal echogenicity	1/41
Preoperative AP pelvic diameter	33 mm*
AP pelvic diameter at follow-up	11 mm*
MAG-3 scan findings	
Preoperative ipsilateral renal function (%)	40%*
Ipsilateral renal function at follow-up (%)	46%*
Obstructive pattern	2/41 (5%)

*p value <0,001.

Table 4. Revision of literature on long term outcomes in vascular relocation in UPJO by crossing vessels.

Author	Year	No. cases	Methods	Mean age at study point (years)	Follow-up (months)
Pesce ⁶	1999	61	US, DTPA	15.5	59 (24-96)
Villemagne ¹¹	2015	70	US, MAG-3	12.7	52 (13-114)
Chiarenza ⁵	2016	35	US, MAG-3	11.5	48 (12-96)
Madec ¹⁰	2016	33	US, Doppler, Blood pressure	15.8	67.2 (31-113)
Present series	2021	41	US, MAG-3, Blood pressure	22.2	152 (72-324)

postoperative analgesic therapy, lower risk of adhesions that makes easier any re-do surgery. The main criticism of the mini-invasive procedure compared to the open one is the greater length of the learning curve necessary to confidently master all laparoscopic surgical times and in particular the suture technique. This problem can be overcome by a complete and gradual training of the specialist, which includes theoretical courses, fellowships in experienced centers, training in laboratory and in the operating room with the help of an experienced laparoscopist.

Conclusions

Our paper establishes that in case of extrinsic-UPJO, VH is very effective to solve the obstruction even in long term follow-up. According a meticulous clinical and instrumental follow-up we can also excluded the stated risk of polar renal hypotrophy and the correlated renal hypertension in adolescent and adult patients. Our results suggest that modified anatomy due to the vascular hitch procedure does not later induce clinically significant vascular injury to the lower pole of kidney during statural growth or lead to hypertension in the long term.

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