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Iatrogenic retractile quadriceps fibrosis within children in Benin: Epidemiological, clinical, therapeutical aspects

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ABSTRACT

Background: In tropical countries, iatrogenic retractile quadriceps fibrosis (IRQF), the cause of walking handicap in children, is often the result of intraquadricipital injection of quinine salts. The aim of this review was to analyse the epidemiological, clinical, therapeutic aspects and outcome of IRQF in children admitted in three hospitals in Benin Republic. **Patients and Methods:** It was a 10-year retrospective, descriptive and analytic survey of IRQF, involving 81 children aged from 8 months to 15 years. Iterative mobilization of the knee (IMK) or modified distal quadriceps plasty by Thompson-Payr's technique (MDQTPT), with a POP on the knee in flexion position, was performed with additional functional rehabilitation. The results were evaluated on knee flexion gain and walking quality. Data were processed using Epi Info 3.2 software. **Results:** Patients' average age was 7.60 years. Children of 6-10 years were most affected; sex ratio was 1.02. Lesions were unilateral (71.6%) and bilateral (28.4%). The knees' stiffness was in flexion (10.57%), rectitude (64.42%) and recurvatum (25%). The amyotrophy of the thigh was found in 79.42 %. The IMK was successful in eight cases (7.69 %) and the MDQTPT was done in 98 cases (94.23%) associated with femoral osteotomy in 13 cases (12.50%). In post-surgical period, skin necrosis and fractures occurred respectively in 15.31% and 5.10%. Results were good in 92.31% of cases. **Conclusions:** IRQF in children do exist in our settings. The treatment that is based on MDQTPT associated to rehabilitation leads to acceptable outcome.

Key words: Child, fibrosis, knee stiffness, plasty, quadriceps

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INTRODUCTION

The iatrogenic retractile quadriceps fibrosis (IRQF) is a cause of physical and social disability in children.[1,2] It is often as a result of intraquadricipital drug injections, especially quinine, mostly used in malaria treatment.[3,4] It may result in fibrosis of quadriceps with adhesion to the femur, causing muscular contracture, shortening extension mechanism with knee stiffness.[4-6] Children with this condition show stiff knee fully extended, causing a limp, awkward sitting and squatting position and could disturb walking.[1,2,7] A variety of surgical techniques such as large detachment of Judet,[7-9] the proximal quadriceps plasty of Sengupta,[10] the distal quadriceps plasty technique of Payr -Thompson (DQTPT)[1,4,11] to reduce muscle contracture are documented.[12,13] Our aim in this survey was to analyse epidemiological, clinical, management and outcome of diagnosed IRQF cases in children.

PATIENTS AND METHODS

This was a retrospective study involving 81 children with 104 cases of IRQF, between January 2000 and December 2009. Medical records and surgical reports were used as data sources and this covered only children under 16. This survey was conducted in three hospitals in Benin.

A standard X-ray of the knee and lateral soft-rays were performed in all patients for knee bones and muscles assessment. In cases, stiffness onset did not exceed 6 months, iterative mobilization of the knee (IMK) was done with general anaesthesia.[14] Beyond 6 months, the surgery was more suitable.[12,13] The MDQTPT lead to lateral and anterior skin incision and extra-articular quadricepsplasty without arthrolysis, based on an extension of the rectus femoris muscle flap with a distal muscle-tendon flap created with a vastus intermedius [Figure 1].The longitudinal incision in the form of an inverse, which basis is oriented to the

patella and the apex to the trochanter, starts at the side of the middle tier of the thigh and stops next to the lateral femoral condyle, bypasses both sides of the patella, stopping next to the medial femoral condyle. The rectus femoris muscle is mobilised and isolated from the vastus muscles and then the pre-patella tendon was cut. Tendon-muscle flap with rectangular form, which size did not exceed 1/3 of the thigh length was performed in the distal vastus intermedius. The electrical incisions bypass the patella on the both sides by cutting the retinaculum of the vastus lateralis and medialis at the femoral condyles level. Dealing with the tensor fascia lata section, the flap is mobilised from muscle vastus intermedius above the periosteum towards the knee so as not to damage the capsule in the quadriceps. At this stage, the knee bends freely. Repeated movements of knee flexion and extension were done of until a maximum flexion was obtained. Suture elongation plasty was performed between the tendon of the rectus femoris and the distal flap vastus intermedius in knee flexion position of 45-60°. Bringing closer sutures of vastus lateralis and medialis muscles are then made on both sides of the edge of the new formation after the muscle plasty [Figure 2]. Wound reparation was performed in two plans. In cases with important recurvatum, femoral osteotomy was realised. A thigh-ankle plaster was carried out for 4 weeks. After the 8th day, wounds were redressed through the window in the plaster. After plaster removal, patient started rehabilitation. In case of lower limbs' length inequality, corrected was provided through sole compensation. Outcome was appreciated at the end of rehabilitation exercises and this was bases on knee flexion gain and quality of walking.[1] The result was rated good if the flexion gained is above 90°, allowing a normal squatting and gait. The result was acceptable if the flexion gained is between 45° and 90° and comes with a normal gait and comfortable sitting position, the result was bad if the flexion gained was under 45° associated with limp walking and uncomfortable sitting. The children were followed-up with variable time ranging from 1 to 10 years. The collected data included patients' age, sex, onset time of knee stiffness, thigh amyotrophy, lower limbs' length, knee stiffness amplitude, associated lesions. The amyotrophy was insignificant if the difference of the thigh circumference is <1.5 cm, moderate if between 1.6 cm and 3 cm, severe if >3 cm. The inequality of low limb length is insignificant if it is <2 cm and important if >2 cm. Data were analysed with Excel 2007 for PC (Microsoft Corporation, USA) and Epi Info 3.2. (Epi Info™ For Windows designed by CDC)

Patients were treated according to MDQTPT.

RESULTS

There were 79 patients in this study, with annual incidence of IRQF ranging from 3 to 11 (mean: 8.1). The mean age of patients was 7.60 years (range: 8 months to 15 years). The age of 6-10 years (38.3%) was the most affected.

Onset of contraction after injection was 7 months (range: 3-15 months). IRQF was unilateral in 58 cases (71.6%) and bilateral in 23 cases (28.4%); the right side was involved in 66 cases (63.46%) while the left side was involved in 38 cases (36.54%). The mean contracture (bending) angle was 1.71° (range: +40° to −70°) [Table 1]. In the case of stiffness, patella could be

Table 1: Knee's stiffness

Knee's position	Maximal value	Mean value	Minimal value	Total	%
Flexion	40°	26.82°	15°	11	10.58
Rectitude	10°	4.5°	0°	67	64.42
Recurvatum	−70°	−29.61°	−15°	26	25.00



Figure 1: Extension of rectus femoris muscle flap with a distal muscle-tendon flap created with a vastus intermedius



Figure 2: View of the new formation after the muscle plasty

mobilised only in 11 cases (10.58%). In a total of 89 cases (85.58%) of fixed patella, 63 cases were as a result of knee stiffness in rectitude position [Figure 3], while 26 cases were due to knee recurvatum [Figure 4]. In four cases (3.85%) of the knee stiffness in a straight position, the patella dislocated whenever patient tried to bend.

Thigh amyotrophy was found in all cases of stiffness in rectitude and recurvatum position (79.42%). In the case of unilateral IRQF, the mean difference between thighs' circumferences was 4.85 cm (range: 2.2-8.5 cm). Lower limbs' length inequality was identified in 43 patients (53.08%) with unilateral IRQF. The mean length difference was 3.6 cm (range: 2.3-12 cm). Bipedal walking was impossible in two patients with severe recurvatum. They moved in quadruped position [Figure 4].

Co-morbid lesions were noticed in four patients and these were necrosis of the femoral head. Femoral head was dislocated in one patient with 12 cm limb length shortening. Bone' bending in case of recurvatum was found on the X-ray images in 23 cases (88.46%). IMK was performed in 11 cases (10.58%) and MDQTPT in 94.23% of cases. Femoral osteotomy was performed in 13 cases (12.5%).

Skin necrosis or sutures anomalies were found in 15 cases (15.31%) after operation. Five cases (5.10%) of femoral metaphyseal fractures occurred during rehabilitation exercises with the arrest of the process and immobilisation in flexion by plaster. In terms of outcome, all patients had a rating of power in the quadriceps muscle above 3 at the end of the rehabilitation. After 5 years follow-up, results were good in 96 cases (92.31%) and fair in eight other cases (7.69%) with recurvatum or treated by osteotomy. MDQTPT results were good in 90 cases (91.84%) and fair in eight cases [Figures 5 and 6]. The average amplitude of flexion



Figure 3: Case of IRQF with unilateral rectitude stiffness



Figure 4: Case of IRQF, patient with severe bilateral recurvatum stiffness



Figure 5: Case of IRQF with limit unilateral rectitude stiffness after MDQTPT



Figure 6: Case of IRQF, patient with severe bilateral recurvatum stiffness after osteotomies and MDQTPT

Table 2: Knee's stiffness correction and result

Surgical procedure	Flexion		Rectitude		Recurvatum		Total	
	Number	Mean flexion	Number	Mean flexion	Number	Mean flexion	Number	%
IMK	6	97°	0	0	0	0	6	5.77
MDQTPT	5	111°	67	90°	18	91.12°	90	86.54
MDQTPT and osteotomies	0	0	0	0	8	82.5°	8	7.69
Total	11	103.36°	67	90°	26	88.46	104	100

IMK: Iterative mobilisation of the knee, MDQTPT: Modified distal quadriceps plasty by Thompson-Payr's technique

obtained following MDQTPT was 90.66° ranging from 70° to 115° [Table 2].

DISCUSSION

As far as epidemiological aspects are concerned, the IRQF, relatively rare disease up to its first description by Hnevkovsky in 1961, has become frequent with the use of intramuscular antibiotics, vaccines and other drugs like quinine.[3,8,11] Since the 1980s, the increasing use of the intravenous route of drug administration instead of the intramuscular route has significantly reduced its incidence in developed countries.[9,11] In opposite, in developing countries, this disease appears more and more.[4,7] The annual incidence of patients in our series (8.1 cases) is lower than that found by Soumah *et al*. (10.2) in Conakry and by Keita *et al*. (14.6) in Bamako and is higher than that found by Onimus *et al*. (2.72) in Central African Republic.[4,5,7] For Burnei *et al*. iatrogenic quadriceps retraction is still an important paediatric issue in countries with poorly organized health-care systems and oversized figures of infantile institutionalization.[11] In such countries, the disease is usually lately diagnosed, when the knee motion is severely limited. In our series, both sexes are affected in the same proportion. Male predominance was clear in the series of Soumah *et al*. (2.17) and Keita *et al*.[4,5] (1.9). Mukherjee and Das reported female predominance with sex ratio of 0.68.[1] The age of 6-10 years was most affected in our series. The age range of 3-7 years was the most affected according to Soumah *et al*.[4] These two results appear similar and might be in relation with the high morbidity in this age group and the importance of walk for parents. The annual incidence of IRQF (10.4) is lower than that found by Burnei *et al*.[11] (18.66%), Keita *et al*.[5] (14.67), is similar to results from the work of Soumah *et al*.[4] (9.2) and is higher than that of Onimus *et al*.[7] (2.7). Cases of unilateral lesions in our series (71.6%) are lower than those revealed by Soumah *et al*.[4] and Onimus *et al*.[7] (100%) and higher than Burnei *et al*.[11] (13%) and Jackson and Hutton[8] (2%). In these last two, the rate of bilateral lesions is higher than ours, 87% and 98% respectively. Regarding the clinical forms of IRQF, the rate of stiffness rectitude was predominant followed by stiffness in flexion and recurvatum. The rate of knee stiffness in rectitude found in our series (64.42%) is similar to that found by Soumah *et al*.[4] (64.13%), higher than that found by Keita *et al*.[5] (45, 45%) and lower than that found by Onimus *et al*.[7] (100%). The rate of stiffness recurvatum in our series (25%) was lower than that found by Mukherjee and Das[1] (29.72%). The rate of flexion stiffness (10.58%) found in our series is lower than that found by Keita (54.55%) and Soumah *et al*.[4] (35.87%). This is the sign of the severity of lesions in our series and the confirmation of the fact that diagnosis was carried out, in most cases, on fixed lesions. Amyotrophy and lower limbs shortness were symptoms of severity. Amyotrophy identified in 79% of cases in our series, was evident in all patients in the series of Soumah and was not evident among patients in the series of Onimus *et al*.[7] Retraction of the quadriceps muscle of which amyotrophy is one of the obvious signs, constitute a limiting factor or a handicap for bipedal walking in children.[2,15] The fact that two patients with recurvatum stiffness were walking in quadruped position is an illustration. Lowers limbs length inequalities and the associated hip diseases in our series were not previously reported. Although new methods of investigation in imaging such as ultrasound[5,6] and magnetic resonance imaging[9,16] provide more information on the local lesion transformations, the standard radiography, the only available method in our context, was sufficient to assess bone deformities. Outcome rate of conservative treatment by IMK in our series was (10.58%). It is lower than that observed by Keita *et al*.[5] (93.18%) and by Jackson and Hutton[8] (43.75%) and higher than Soumah *et al*.[4] (6.52%). The rate of surgery in our series (94.23%) is similar to that reported by Soumah *et al*.[4] (93.48%). This rate is lower than that of Burnei's *et al*. (100%)[11] and higher than those of Jackson and Hutton[8] (56.25%) and Onimus *et al*.[7] (72.73%). According to Burnei *et al*.[11] effective prevention was set into medical practice before there was a consensual decision for surgical treatment. Regarding quadriceps retraction treatment, some authors prefer surgery with limited dissection while others choose a more extensive dissection in order to visualise all elements involved in fibrosis.

Soumah *et al*.[4] considered that modified technique of Payr-Thompson based on an extension in Z, V or Y of the tendon reduces post-operative adhesions, which often occur at the major muscle trauma interventions in children as in Judet's techniques. Modification performed with lateral incision, which offers a better view of all the distal quadriceps according to Mukherjee and Das,[1] by an extension of semi-circular incision above the two femoral condyles prevents from damage to nerves, vessels and periosteum. With this modification, all surgical manipulations remain extra-articular and there is an assurance of the integrity of the bursa and articular surfaces. Pathogenesis research proved that the vastus intermedius is the most injured, followed by the vastus lateralis muscle and the rectus femoris.[1,6,17] The muscles, vastus intermedius and rectus femoris are in the same axis and their role is complementary in knee extension. The high section of the vastus intermedius (Judet), the V or Z lengthening of the quadriceps tendon (Payr-Thompson), induce at the same time the elongation of the length of these two muscles, which led to the occurrence of poor extension that was incriminated in follow-up by authors.[7,8] In this modified technique, the mobilisation phase of the rectus femoris and the distal flap from the vastus intermedius is a bit similar to the detachment method of Judet. After the plasty of the quadriceps muscle, the transformed muscle could be identified as "triceps femoris" in anatomical point of view. The rate of complications such as greenstick metaphyseal femoral fracture found in half of cases with conservative treatment is higher than that observed by Jackson and Hutton[8] (14.3%). This could be explained by the fact that these cases of IRQF were wrongly considered as recent condition. The rate of skin necrosis or sutures anomalies observed in this series (15.31%) in the post-operative period is higher than that found by Mukherjee and Das[1] and Burnei *et al*.[11] (12.8%). Cases of hematoma have been reported by Burnei *et al*.[11] (1.1%) and Onimus *et al*.[7] (18.75). We did not found such events in this series. The rate of good results (91.84%), which is the expression of an average knee flexion gained 90.66 ° with a normal active extension is greater than the rate obtained by Soumah *et al*.[4] (56.5%) and Jackson and Hutton[8] (57.5%). The mean flexion gain was on the other hand higher than the results obtained by Burnei *et al*.[11] in his two different groups, using the technique of Judet (85°) and Payr-Thompson (78°). Strict physiotherapy has allowed a significant increase in the amplitude of bending to the operated knee and increased mass and muscle strength.[18] The younger patients had a better recovery in our series, as noticed by Mukherjee and Das[1] The good results obtained with the use of this modified technique of distal Payr-Thompson's quadricepsplasty, allow us to recommend it for the surgery of fixed IRQF. Nevertheless prevention must be promoted in order to avoid new cases of IRQF in children. This includes intramuscular injection prohibition in children with preference to oral or intravenous routes for drugs administration.[2,5,7]

CONCLUSION

IRQF in children is common in our hospital. Treatment focuses on MDQTPT associated with strict physiotherapy allows a satisfactory rehabilitation of the patient. Intra-quadricipital drug injection in general and the quinine salts solutions in particular, should be avoided/banned in children as much as possible.

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