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ORIGINAL STUDY

Cozen's phenomenon : state-of-the-art management taking the advantages of recent advances in growth modulation with critical review'

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Cozen's phenomenon is an uncommon condition with an unclear protocol for management. The purpose of this study was to emphasize a reliable plan for the treatment of these cases. Case series of 14 patients fulfilling the determined inclusion criteria were treated using 8-plate hemiepiphyseodesis. Their mean age at the time of the operation was (4 y & 7m). The mean preoperative anatomical femoro-tibial valgus angle was 22°. Overcorrection (~ 5°) was obtained in all the cases at the end of treatment after a mean period of 15 months. Although two cases showed early rebound at the 8th month of follow up, it was temporary and all the cases fortunately after a mean follow up period of 28 months showed complete resolution. 8-plate is a simple, effective, and safe technique that works successfully when used properly in selected cases of Cozen's phenomenon.

Keywords : Cozen ; genu valgum ; hemiepiphyseodesis ; 8 plate ; tension band.

INTRODUCTION

"The propensity of the non-displaced proximal tibial metaphyseal fractures to develop delayed-onset progressive tibial valgus deformity in children was first reported by Dr. Lewis N. Cozen, in 1953 *(5,8,10-12)*. Therefore, it righteously carried his name, "Cozen's Phenomenon". It is an uncommon condition with unclear protocol for management. Formerly, orthopedists recommended intelligent negligence, also others documented its failure *(5,1,32,34,36,44,45,49,50)*. High tibial osteotomies (HTO) was considered for treatment however many issues were recorded due to high recurrence rate *(11)*, the likelihood of developing secondary deformities, longer hospital stay, as well as patient inconvenience owing to the presence of a considerable postoperative pain, cast immobilization, and delayed weight bearing *(5,6,11,28,32,37,39 40)*.

The aim of this work is to emphasize a reliable plan for treatment of cases with Cozen's phenomenon taking advantage in the recent advances of growth modulation on temporal bases using tension band 8-plate.

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Conflicts of interest: none

PATIENTS AND METHODS

Fifteen legs in 14 patients (8 boys, 6 girls) with Cozen's phenomenon who presented to the orthopaedic department between January 2011 and February 2014 and fulfilled the inclusion criteria were included in this prospective case series study. The study was approved by the local ethical committee (General Organization of Teaching Hospitals and Institutes, Research Ethical Committee) and was conducted in accordance with the Declaration of Helsinki and Ethical Guidelines for Epidemiological Research. Informed written consents were obtained from all patients and their guardians before participation in the study. Eight cases were right sided, 5 were left sided and one case had bilateral affection. The mean age of the patients at the time of injury was (3 y & 3m), (range, 1y & 8m-6 y, SD 1y & 5m). The mean time passed post injury till genu valgum first observed for the included cases was 4 months, (range, 2-7m, SD 1.6). The mean age of the patients at the time of the operation was (4 y & 7m), (range, 3y & 6m-7y &7m, SD 1y & 3m). The mean preoperative anatomical femoro-tibial valgus angle (aFTA) was 22° (range, 15-30, SD 5) (Table I). All the cases gave a history of domestic trauma except in 2 cases (no.7 & 13) that developed the deformity after having high tibial osteotomy as a treatment for genu varus deformity that the authors metaphorically gave them the name "***iatrogenic*** Cozen's phenomenon" (Fig. 1). *The inclusion criteria* were : (1) cases presented initially with severe valgus with (aFTA) is > 15° ; (2) rapid progression of valgus deformity during 6 months of follow up ; (3) development of compensatory secondary deformities of the distal femur or distal

Table I. — Summary of the cases :

Case	Gender	Age at injury (y+m)	Side	Time passed post injury till G. valgum 1st observed (m)	Age at surgery (y+m)	aFTA at time of treatment	Total duration of hemiepi-physeodesis (m)	Age at last FU (y+m)	FU period (y+m)	Comment	Final result
1	M	3	R	6	4+6	23°	18	7+10	2		Resolved
2	M	5	L	4	6+10	17°	12	10+4	2+6		Resolved
3	M	1+8	R	3	3+6	20°	13	6	2+5	Epiphyseal screw back out that wase revises	Resolved
4	M	2	R	5	3+8	20°	12	6+8	2		Resolved
5	F	6	R	7	7+7	17°	17	11+6	2+6	Rebound occurred *	Resolved
6	F	2	R	2	3+10	27°	16	7+2	2		Resolved
7	F	3+4	Bil	6	5	R: 15° L: 25°	13 18	9	2+11 2+6	Iatrogenic Bil Cozen's phenomenon **	Resolved Resolved
8	F	3	R	3	4+9	30°	24	8+10	2+1		Resolved
9	M	2	L	3	3+6	22°	16	7+6	2+8	Rapid valgus progression	Resolved
10	F	3+6	L	5	4+11	17°	13	8+6	2+6		Resolved
11	M	6	L	3	7+6	17°	12	11	2+6		Resolved
12	M	2	L	3	3+6	30°	16	6+10	2	Rapid valgus progression	Resolved
13	M	3	R	4	4	25°	16	8	2+8	Iatrogenic Cozen's phenomenon **	Resolved
14	F	3	R	2	4+2	18°	12	8	2+10	Rebound occurred *	Resolved

Y : year ; M : month ; aFTA : anatomical femoro-tibial angle ; * : Rebound occurred, reaching its maximum (5° valgum) compared to the healthy side on the 8th month after stoppage of growth modulation. Improved markedly at the end of follow up. ** : Iatrogenic valgus after corrective high (metaphyseal) tibial osteotomy for treatment of genu varum deformity.

Fig. 1. — An intraoperative image intensifier view showing synchronous 8 plate hemiepiphyseodesis of a case of bilateral "iatrogenic Cozen's phenomenon" (case no. 7 in the table).

Fig. 2. — x-ray after extraction of the metaphyseal screw after attaining slight overcorrection in relation to the other limb.

tibia ; (4) persistence of the deformity beyond 2-year follow-up ; and (5) cases older than 4 years. *The exclusion criteria* were : (1) cases younger than 2 years and (2) cases showing improvement on follow-up.

Preoperatively, the degree of valgus deformity was measured using the aTFA on a long lower limb radiograph. All cases were treated using the same strategy of proximal medial tibial temporary 8-plate hemi-epiphysiodesis applying the same operative technique described by Stevens PM *(40)* with the exception that non-cannulated screws were used. Postoperatively, patients may mobilize quickly without restrictions as tolerated. Follow up was every 3 months for clinical evaluation of correction of the angulation. A follow up standing anteroposterior radiograph is taken when the legs are clinically straight. In the first 5 cases, Extraction of the metaphyseal screw took place when the aFTA was intentionally slightly overcorrected as regard to the other side by ~ 5° (Fig. 2). The remaining hardware was then extracted after 6 months if no rebound occurred ; otherwise this metaphyseal screw was planned for re-insertion again. This was not needed in any of these cases. So, in the remaining cases, all the hardware was removed in one setting. The minimum follow-up period was 2 years after cessation of hemiepiphyseodesis.

For the statistical analyses, IBM SPSS v.22 program (SPSS Inc., Chicago, Illinois, USA) was used. Descriptive analysis, independent samples *t*-test, and Spearman correlation were used for analyzing the results. The significance level was set at $p < 0.05$.

RESULTS

Overcorrection (~ 5°) was obtained in all the cases before termination of the growth modulation. The mean period of the temporary hemiepiphyseodesis was 15 months (range, 12-24m, SD 3). The mean age at the last follow-up 8y & 4m (range, 6y-11y & 6m, SD 19.6m). The mean follow-up period was 28 months (range, 24-34m, SD 3.7 m). All cases resolved at the end of the follow-up (Fig. 3). There were no recognized neurologic or vascular complications. No iatrogenic affection to the proximal tibial physis was seen at last follow up in any patient. In 2 cases (cases no. 5 & 14) a rebound was observed, reached its maximum 5° valgus in excess of the healthy side on the 8th month after termination of the growth modulation. they showed spontaneous great improvement at the end of their follow up. One case (case no.3) showed epiphyseal screw back out on the 6th months follow up. It was revised and the case continued smoothly until the

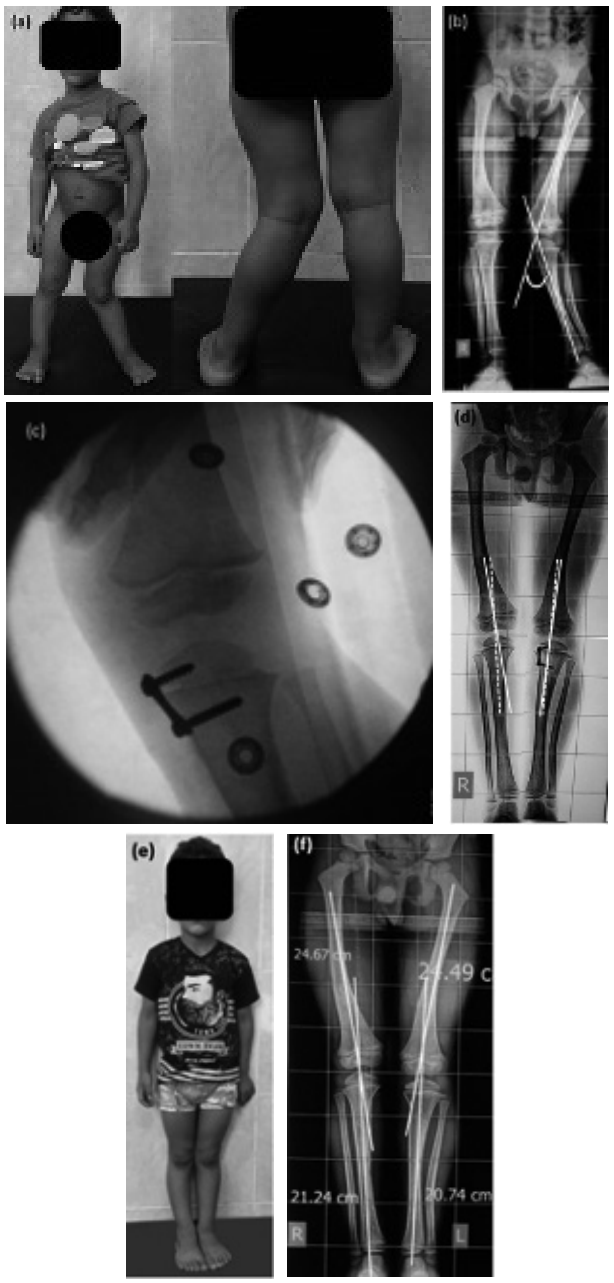

Fig. 3. — (a) A 3 ½-year-old male child at the time of surgery with a left Cozen's phenomenon (case no. 12 in the table). (b) The preoperative scanogram showing an aFTA valgus angle 30°. (c) An intraoperative image intensifier view showing the 8-plate bridging the upper medial tibial physis. (d) Pre-extraction scanogram after 16 months showing overcorrection compared to the healthy side. (e & f) last follow up at 2 years after extraction showing no loss of correction with tibial shortening less than 1 cm.

end of treatment with the maintenance of correction at the end of follow up. No LLD was found at the end of follow up except in two cases who showed ipsilateral tibial shortening, but fortunately, it was less than 1 cm.

There was a nonsignificant correlation between age at the time of surgery and the total duration of the growth modulation (r = .064, P = .829), but there was a significant correlation between the pre-operative aFTA and the total duration of the growth modulation (r = .625, P = .013). There was no statistically significant difference of the total duration of growth modulation between the 2 genders (t = -.794, P = .442).

DISCUSSION

Late-onset tibia vulga deformity is a well-known disorder that develops insidiously after undisplaced proximal tibial metaphysis fractures in children *(10,11)*. It may start and progress as early as during fracture healing where it appears on cast removal (6-8 weeks) or as late as 12 months or more after the initial injury. It was mentioned that the greatest progression takes place within the first few months. And after approximately 1 year it nearly stops progressing and starts correction *(2,5,6,11,12,19,22,32,36)*. Nenopoulo, S. *el. al.* in 2007, showed that almost all children with this type of fracture developed late tibia vulga, where (90.3%) of their case series (39 patients) developed this phenomenon *(32)*.

A multitude of etiological factors for this phenomenon had been postulated in the literature. But the actual cause had not been settled yet. The theory of the "uneven overgrowth phenomenon" is the most accepted one, where it owed this to the stimulated growth of the proximal tibial physis as a whole by the fracture in the presence of a tethering effect of the fibula laterally *(1,8,11,16,21,36,49)*. This will lead to differential over-lengthening of the medial side of the tibia relative to its lateral one. Two other accepted hypothesized etiological causes are either that of the soft tissue incarcerated at the medial end of the fracture site mimicking medial opening wedge *(11,19,24,27,33,45,46)* or that of the proximal medial tibial metaphyseal hyperaemia

which leads to a sectorial growth stimulation of its nearby medial hemi-physis *(16,17,21,49)*. A fourth and least accepted mentioned cause was that owing this phenomenon to the plastic deformation resulting either from the initial or a later loss of reduction *(10,34)*. In this work, we inclined towards the first hypothesis on which we built our treatment strategy, which is the growth modulation.

Earlier literature on Cozen's phenomenon was unfortunately as case reports and for the most part, an audit made in respect of authors' personal experiences that lacked for the criteria for either recognizing children at risk of developing this deformity or predicting its spontaneous correction. *(5,6,11,12,23,31,34,36,45,46)*. This made this condition to be managed on anecdotal evidence.

There is no general agreement in the literature about the incidence and degree of valgus deformity that may follow Cozen's fracture. Few studies have addressed the untreated course of this deformity as it occurs after trauma. Salter *el. al.* in 1972 found no spontaneous correction of post-traumatic tibia valga in their series of twenty-one children *(11,35)*. Müller I *el. al.* in 2002 concluded that partial remodeling is seen only in children up to 5 years old *(31)*. Visser *el. al.* in 1982 *(45)*, Skak SV in 1982 *(36)*, Robert M *el. al.* in 1984 *(34)* and Zionts L E *el. al.* in 1986 *(50)*, recommended intelligent neglect for this deformity as it did resolve over time, additionally, they observed that correction took place minimally from the proximal and mainly from the distal tibial growth plate producing a radiological S-shaped tibia with a skew mechanical tibial axis. Again, Zionts, L. E. with Tuten, R. *el. al.* in 1999 re-evaluated their previous 7 cases included earlier in their initial publication in 1982 on a long term follow up basis where they observed full clinical alignment just by intelligent neglect policy but with a resultant radiological S-Shaped tibia *(44)*. Although Jackson, D. *el. al.* in 1971 came to the conclusion that correction may be spontaneous, they added that once genu valgus exceeds 15 degrees, a complete correction will not occur *(19)*. A finding confirmed also by Ippolito E *el. al.* in 1984 *(18)*. Nenopoulos, S. *et. al.* in 2007 re-evaluated 11 patients with an angulation >5° after 7 years from the initial injury. They observed non-remodeling in (20%), Partial remodeling in (55%) and total remodeling in (25%) of cases *(32)*. Finally, remodeling is more expected in children less than 4 years old *(5,18,19)*.

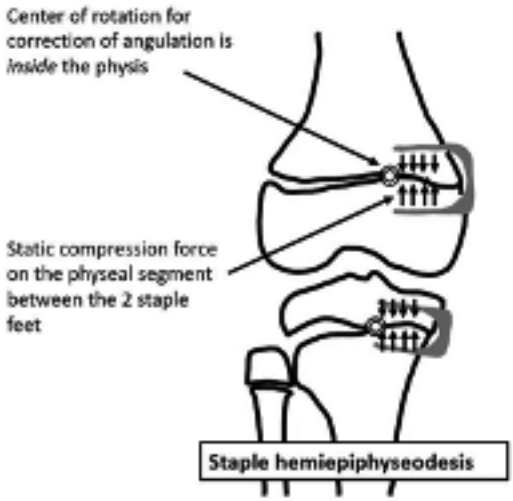

Fig. 4. — A diagram shows how the staple works. The center of rotation for correction of angulation is inside the physis. There is a static compression force on a significant portion of the physis.

Although intelligent negligence was recommended by some authors as a line of treatment, especially for children younger than 4 years old, unfortunately, remodeling reported to start as late as 3 years' post-injury and lasts up to 15 years *(5,11,19,29,32,34,36,44,45,50)*. All these papers lacked clear information regarding the way of corrective remodeling, a clear plan of management, and indications for interference. It was found that intelligent negligence is practically not accepted for many reasons : (1) this deformity is very alarming for the parents *(5,19,32)* ; (2) known lack of satisfactory remodeling power of the proximal tibial physis *(32,45)* ; (3) valgus deformity, in contrarily to varus, is intolerable by the patient, increases with weight bearing, and only partially corrected with age *(31,47)* ; and (4) High possibility of developing a secondary reversed compensatory deformities either below through the distal tibial physis resulting in an "S-shaped tibia" or above through the distal femoral physis resulting in a distal femoral varus (false correction) *(32,34,36,45)*.

Growth modulation on a temporary basis was first applied in human beings by Blount WP and Clarke GR in the late 1940s by using staples. It

works primarily by exerting a *static compression force within* the physis segment bracketed by its 2 prongs. This leads to sectorial physeal arrest of this segment and consequently, the center of rotation of correction of angulation will be located *inside* the physis *(3,4,15)* (Fig. 4). After this technique enjoyed several decades of popularity, it fell into disfavor in the 1980s due to the many complications reported as staples migration, back out, breakage, rebound phenomenon, the difficulty of retrieval, and the most important shortcoming was the likelihood of physeal bar formation causing permanent growth arrest. This was explained by the long-standing inhibitory compression forces exerted on the physis between the two staples' prongs *(7,9,14,25,51)*. Consequently, stapling had considered problematic in young children *(3,7,15,30,38,48)*. In 2004, Stevens, P. M. advented his extraphyseal, extraperiosteal, flexible, dynamic tethering non-locking 8-plate guided growth system. It is a *tension-band-like plating* that acts as an external hinge ; without any compression forces exerted upon any part of the physis**.** Consequently, the center of rotation of correction of angulation will be entirely located *outside* the physis *(7,13,39)*. So, as the physis grows the screws toggle in the plate leading to gradual correction that takes place all over the whole diameter of the physis *(7,39)* (Fig. 5). This reduces the concern of permanent physeal damage, bar formation or rebound phenomenon *(2,30,41)*. The recorded safety of 8-plate hemiepiphyseodesis as well as the known healthy nature of the proximal tibial physis that still has growth potential in these cases, makes the procedure an appealing line of management for as younger cases as 2 years old *(7,13,15,20,26,40,43,48)*.

To date, the published studies on using 8 plate for treating Cozen's phenomenon were as case reports. Visser JD and Veldhuizen AG in 1982 were the first reporters of using temporary hemiepiphyseodesis in 4 cases with Cozen's phenomenon using staples without mentioning any details of the cases. Henceforward, Stevens PM *el. al.* in 2006 reported 11 cases treated mainly by staples and little with 8-plating without giving sufficient details. Recurrence was observed in six of their cases that required repeated stapling

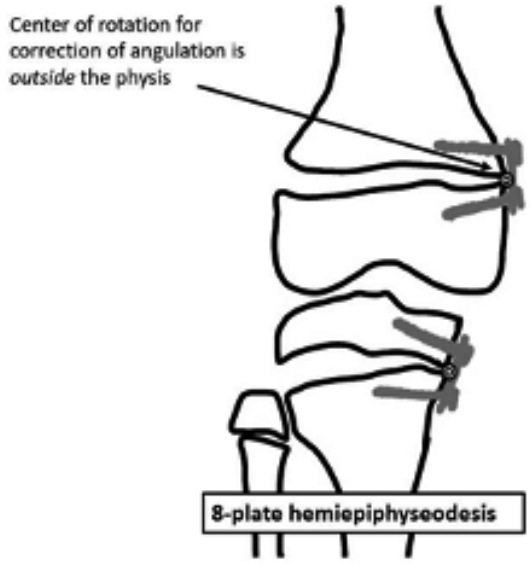

Fig. 5. — A diagram shows how the 8-plate works. The center of rotation for correction of angulation is completely outside the physis. So, this plate will act as an outside hinge without performing static compression on any part of the physis.

with a successful final outcome. Since then, the 8 plate hemiepiphysiodesis has gained widespread popularity *(42)*. Recently, in 2013, Dorman S, *el. al.* published a case report for a four-year-old boy with a right Cozen' phenomenon after 20-months post-injury treated by 8-plate hemi-epiphyseodesis. The deformity corrected successfully after 19 months and the plate was removed 2 months later *(12)*. In this study, it was tried to discuss the use of this plate on a homogenous group of cozen's phenomenon with clear selection criteria. And they all reported a final successful outcome at the end of follow up. The adverse effects reported during treatment were all reversible.

Although the deformities of 3 included legs in this study were iatrogenic as late sequelae to a previous high tibial (metaphyseal) osteotomies for treatment of genu varum "iatrogenic Cozen's phenomenon", studies are needed to further confirm these findings. However, until this concept can be affirmed in a larger study that achieves general clinical acceptance, the authors urge orthopaedic surgeons to do this osteotomy as more distally as at the proximal third tibia (diaphyseal) rather than the metaphyseal one. This hypothesis was found to

go with many published experimental and clinical studies *(1,33,37)*.

It is very important to weigh early surgical interference against waiting for spontaneous resolution for each individual case based on the progress and degree of the deformity together with the development of secondary deformities, as mentioned earlier here under patient selection.

A shortcoming of this work was the relatively small number of the case series, but this was due to the relatively uncommon nature of this phenomenon. But follow up of these children until maturity to assure maintenance of correction during growth is recommended.

Conclusions : The 8-plate hemiepiphyseodesis is a simple, effective, and safe technique that works successfully when used properly in selected cases of Cozen's phenomenon.

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