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Angular Improvement with Bracing Over 7 Years in Anterolateral Tibial Bowing without Neurofibromatosis Type 1: A Case Report

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Learning Point of the Article:

In anterolateral tibial bowing without neurofibromatosis type 1, bracing may accompany improvement of the angular deformity while limb length inequality progresses independently and requires its own operative plan.

Abstract

Introduction: Congenital anterolateral bowing of the tibia is a rare dysplasia of the tibia that is strongly associated with neurofibromatosis type 1 (Nf1). It produces two deformities, angulation and limb shortening, which are usually reported together and corrected surgically. We report a child without NF1, managed non-operatively, in whom these two courses diverged: The angular deformity improved while the shortening did not. To the best of our knowledge, this dissociation has not previously been documented over more than7 years of continuous non-operative management in a prenatally detected case without a genetic association.

Case Report: An 8-year-old Hispanic boy had right-sided anterolateral bowing of the tibia detected on obstetric ultrasound at 27 weeks of gestation. Genetic testing showed only variants of uncertain significance, and no systemic condition, cutaneous stigma, or family history of NF1 was found. Serial radiographs demonstrated mid-diaphyseal bowing of the tibia with a patent medullary canal and no fracture or pseudarthrosis over more than 7 years, corresponding to Paley type 1. Magnetic resonance imaging showed normal ossification without pathological periosteum. He was managed with a right total contact orthosis and a graduated shoe lift. Tibial angulation improved radiographically and in the appearance of the limb, without a bone defect. Limb length discrepancy was unaffected by bracing, measuring 4.4 cm at latest follow-up, and corrective surgery is now warranted.

Conclusion: Angular deformity in anterolateral bowing of the tibia can be managed conservatively. Limb length inequality can follow an independent course, because the shortening arises from growth inhibition in the affected segment rather than from the angulation. An intact and straightening tibia should therefore not be read as protection against shortening. Limb length warrants projection to skeletal maturity and a separate operative plan from the first consultation, a lesson that applies to every clinician who follows a bowed tibia to maturity.

Keywords: Anterolateral tibial bowing, congenital pseudarthrosis of the tibia, neurofibromatosis type 1, limb length discrepancy, non-operative management, case report.

Introduction

Congenital tibial bowing is a spectrum of deformities present at birth, classified by the direction of angulation into anterolateral, posteromedial, and anteromedial patterns, each with a distinct natural history [1,2,3]. The anterolateral pattern is the least benign of the three. Anterolateral tibial bowing (ALTB) is not a positional deformity but a localized dysplasia in which the periosteum is the primary site of disease and the osseous

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abnormality is secondary. Fibrous hamartoma replaces healthy periosteum, increasing osteoclasis and reducing osteogenesis. The affected segment therefore bows progressively and, in most reported series, eventually fractures. This dysplasia is encountered most often in the setting of neurofibromatosis type 1 (NF1).

NF1 affects approximately one in 2,662 live births [4], and the same periosteal disease underlies its tibial manifestations. Loss of neurofibromin activates rat sarcoma/mitogen-activated protein kinase signaling, impairs osteoblast maturation, and accelerates osteoclast-mediated resorption, which accounts for both the progressive deformity and the failure of union [5, 6]. The end point of that failure is congenital pseudarthrosis of the tibia (CPT), which develops in 2–6% of children with NF1 [1]. CPT follows a fracture through the dysplastic segment that does not unite, so that a false joint forms at the apex. Preventing this outcome is the object of treatment in ALTB [7].

Bowing without a genetic association is frequently described as rare. In one consecutive operative series of patients with ALTB, approximately 30% carried no genetic diagnosis [8]. In a separate cohort of children with ALTB, nine of 40 (22.5%) were NF1 negative [9].

The majority of these patients require surgical management, and few cases have been reported to resolve without operation. In a series of 43 children with congenital anterolateral bowing, five (12%) corrected without progressing to fracture [10]. Among the unilateral cases, limb length inequality was the sole residual deformity at a mean follow-up of 58 months. Angulation and shortening therefore do not follow the same course. We report a child with prenatally detected ALTB and no genetic association in whom prolonged bracing accompanied improvement of the angular deformity, while limb length inequality progressed independently.

Case Report

An 8-year-old Hispanic boy has been followed in our pediatric orthopedic clinic since 1 year of age. He was referred with congenital bowing of the tibia detected on a level II obstetric sonogram at 27 weeks of gestation, which showed isolated right tibial bowing with otherwise normal fetal anatomy.

The mother tested positive for Zika virus in the second trimester. Serial fetal assessment every 3–4 weeks showed growth appropriate for gestational age and normal middle cerebral artery Doppler studies. No microcephaly, intracranial calcification, ventriculomegaly, or arthrogryposis was identified prenatally or on postnatal examination and neurodevelopmental follow-up, and the tibial deformity remained the sole structural abnormality. No family history of NF1 was reported.

At 3 years of age, examination showed symmetric hip abduction, normal femoral anteversion, moderate internal tibial torsion, right anterolateral tibial deformity, and a clinically estimated limb length discrepancy of 2 cm. There were no cafe-au-lait macules, axillary or inguinal freckling, cutaneous neurofibromas, or Lisch nodules. Chromosomal microarray identified only a 360 kb gain of uncertain significance at 7p15.3 involving DNAH11 and CDCA7L, a locus with no established phenotype in the heterozygous state and no relation to skeletal dysplasia; no copy number abnormality of NF1 (17q11.2) was found.

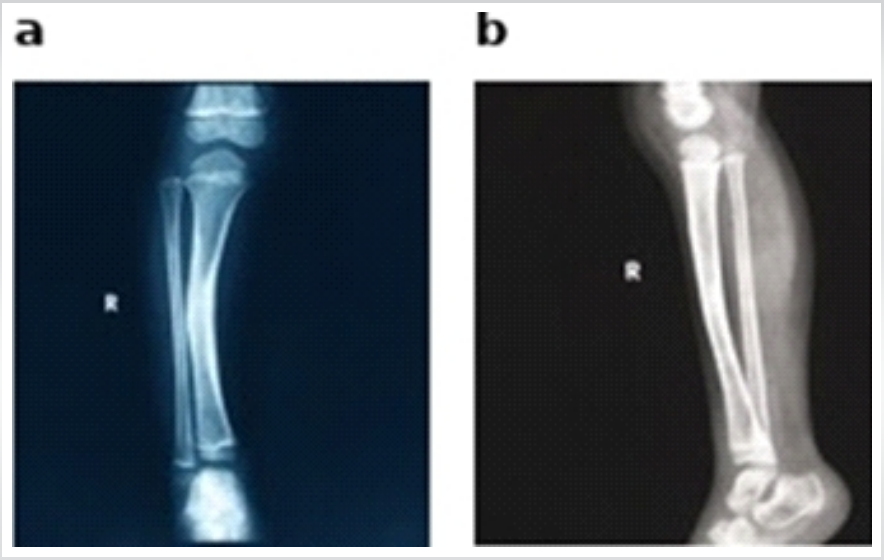

Figure 1: Anteroposterior and lateral radiographs of the right tibia and fibula. Anteroposterior (a) and lateral (b) radiographs of the right tibia and fibula obtained in 2021, at approximately 4 years of age, demonstrating anterolateral bowing of the tibia at the mid-diaphysis without fracture. The characteristic anterior and lateral curvature is present with a patent medullary canal, without hourglass tapering, cystic change, or cortical breach, corresponding to Paley type 1.

Radiographs demonstrated anterolateral bowing at the mid-diaphysis of the tibia without fracture (Fig. 1). The medullary canal remained patent at the apex, without hourglass constriction, cystic change, or cortical breach, corresponding to Paley type 1 [7]. Magnetic resonance imaging showed normal ossification for age, without discrete lesions, cortical disruption, pathological periosteum, or pseudarthrosis. No low-density line, the linear intracortical radiolucency associated with impending fracture, was identified on any radiograph obtained during the period of follow-up.

He was managed non-operatively throughout with a right total contact orthosis worn continuously and a shoe lift. Serial radiographs

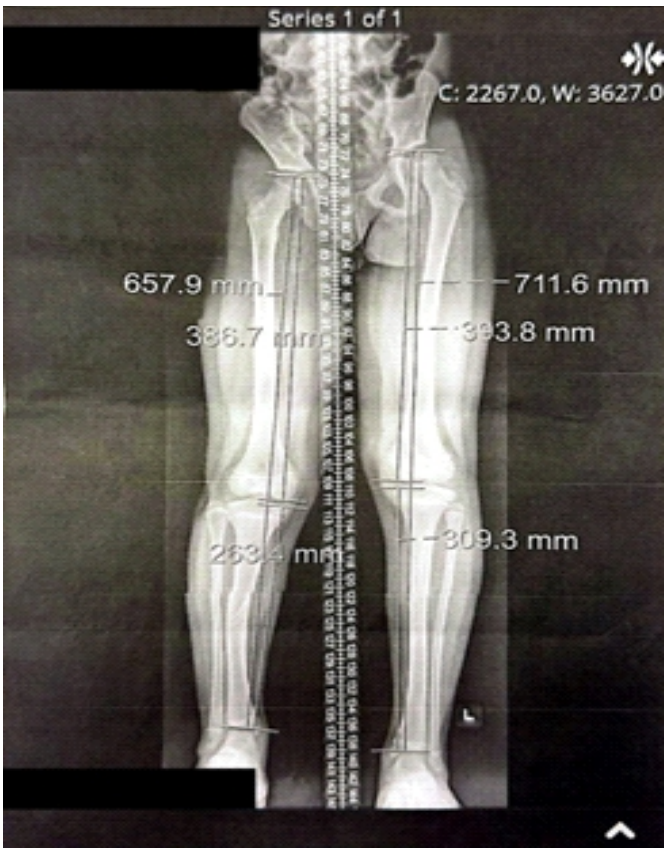

Figure 2: Standing lower extremity scanogram after more than 7 years of bracing. Standing anteroposterior scanogram of both lower extremities obtained in February 2026, after more than 7 years of total contact orthosis and shoe-lift treatment, demonstrating improvement in tibial bowing on the right together with the residual limb length discrepancy. Segmental measurements are displayed on the image. Bone density is normal and there is no fracture. Patient identifiers have been removed from the image.

showed progressive improvement in anterior angulation without bone defect, and a standing lower extremity scanogram at latest follow-up confirmed the improved alignment (Fig. 2). The clinical appearance of the limb improved markedly in the assessment of the treating surgeons and the family. He remains ambulatory and pain-free, with no fracture, pseudarthrosis, or ankle valgus. Limb length discrepancy measured 4.4 cm at latest follow-up and corrective surgery is now warranted.

Discussion

We report an 8-year-old boy with congenital anterolateral bowing of the tibia detected on a second-trimester obstetric sonogram, in whom no genetic association could be identified, and who was managed non-operatively from infancy with a total contact orthosis and a graduated shoe lift. Over more than 7 years of follow-up, the tibia did not fracture, no low-density line appeared on any radiograph, and the angular deformity improved both radiographically and esthetically. The limb length inequality followed an independent course, progressing from 2 cm at presentation to 4.4 cm despite uninterrupted bracing, and now requires an operative plan of its own. The interest of this case lies in that dissociation, since a tibia that remained intact and straightened over 7 years of growth offered no protection against shortening of the affected segment.

The best comparable entity previously reported is a group of ten patients with bowing accompanied by ipsilateral duplication of the hallux without NF1 association [11,12]. The bow resolved spontaneously in all of them, and none progressed to pseudarthrosis, yet every child developed a limb length discrepancy and several required lengthening or contralateral epiphysiodesis to address it [11]. Angulation and shortening therefore diverged in that group as they did in our patient. However, our patient does not represent the aforementioned condition, since that condition is recognizable at birth by the duplicated toe. Other non-syndromic presentations reinforce the heterogeneity of the entity: The delta tibia, a triangular anterolateral angulation with a thickened concave cortex, follows a comparatively benign course [13], and isolated cases of anterolateral bowing without NF1 continue to be reported with variable outcomes [14].

A comparable natural history to that of our patient is described in the series of Tuncay et al. [10]. Of 43 children evaluated for anterolateral bowing, five corrected spontaneously without fracture, none of whom had NF1 or other congenital anomalies. Limb length discrepancy was their only residual deformity and reached 4.8 cm at a mean follow-up of 58 months.

Anterolateral bowing in children without NF1 has rarely been described. A negative genetic workup is often taken to be reassuring in itself, but the available evidence does not support this. Comparisons between patients with and without NF1 have shown no difference in the onset of bowing or fracture, tissue pathology, or union after surgery [15,16]. Paley has summarized the outcomes as equivalent in both groups [7]. The distinction is also less clean than it appears, since somatic NF1 inactivation has been identified in the periosteum of 21% of patients who did not meet clinical criteria for neurofibromatosis [17]. Tian et al. studied 40 children with bowing and found no association between NF1 status and the time to fracture, with five of the nine NF1-negative children going on to fracture [9]. A negative workup therefore carries no prognostic weight of its own, and the dysplastic tibia can fracture and the deformity progress whether or not NF1 is present.

Management of the intact bowed tibia aims to prevent the fracture that converts a manageable deformity into pseudarthrosis. Bracing remains first line, while prophylactic bypass grafting [18] and distal tibial guided growth [19, 20] are reserved for tibiae that deteriorate under observation; once

fracture occurs, the reconstructive burden rises steeply [8,21,22]. Our patient required none of these, since his tibia straightened under bracing and remained intact. His remaining problem is length, and the operation therefore belongs on the sound limb. Corrective surgery has been planned, and the choice between contralateral epiphysiodesis and ipsilateral lengthening depends on the discrepancy projected to skeletal maturity [23].

This report carries the limitations of a single case. No causal claim can be made for the orthosis, since spontaneous correction under observation alone is well documented [10] and prospective evidence for the effectiveness of bracing is lacking [5], so what we observed may represent natural history rather than treatment effect. Angular correction was judged by the treating surgeons and the family rather than measured by blinded observers. The patient has not reached skeletal maturity, so neither the durability of the correction, the final inequality, nor the adequacy of the planned procedure can yet be stated.

Conclusion

In this child with ALTB and no identified genetic association, non-operative management was accompanied by improvement of the angular deformity and freedom from fracture through 7 years of skeletal growth, while limb length inequality progressed independently. Bracing acted on the bow but not on the shortening, and correction of the discrepancy now requires a separate operation. Whether bracing or early operative correction is preferable in the pre-fracture tibia remains unsettled, and a single case does not resolve it.

Clinical Message

Anterolateral tibial bowing produces two deformities with different mechanisms and different responses to treatment. Bracing may protect the dysplastic segment and allow the bow to improve, but it has no effect on limb shortening, which arises from growth inhibition in the affected segment. An intact, straightening tibia should not be taken as evidence that secondary changes are being averted. Limb length should be projected to skeletal maturity from the first consultation and planned for as a separate procedure.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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