Trevor’s disease: up-to-date review of the literature with case series
Kemal Gökkuş, Halil Atmaca, Ergin Sagtas, Murat Saylik and Ahmet T. Aydin

Trevor’s disease, also known as dysplasia epiphysealis hemimelica, is a rare nonhereditary skeletal development disorder that affects epiphyses. This type of dysplastic lesion was first reported by Mouchet and Berlot in 1926 under the name ‘tarsomegaly’. The main aim of this study is to raise awareness of Trevor’s disease among orthopedic surgeons and underline some important aspects of treatment by a detailed presentation of four different possible manifestations of the disease. Four different treatment methods were used on four different patients (three localized in hindfoot ankle region and one classic Trevor’s disease case). Treatment methods, localization of the sides involved, different characteristics of entire lower extremity, asymmetry, distal femoral lateral epiphysis involvement, and hip involvement were analyzed thoroughly and the results were compared with those found in the most recent literature. Of our four patients, three were localized (hind foot ankle) cases and one was a classic dysplasia epiphysealis hemimelica with hemimelic distribution of the entire lower extremity. We used arthroscopic resection, observation, excision, and temporary hemiepiphysiodesis treatment methods in each of our cases. Clinical follow-up results were reported to be between good and excellent. In sum, our opinion is that the treatment for this condition should be customized according to lesion localization and lesion size. Majority of cases with ankle involvement show good prognosis following excision. Observation is also an alternative in patients who refuse surgery. If an intra-articular lesion is present, the surgeon should perform an arthroscopy for assessment of lesion surface. If the lesion is adapted to the joint curvature, it should be left alone and hemiepiphysiodesis should be considered for correction. The most risky involvements that are related to deformities and limb-length discrepancies are the hip and the knee. This is usually the result of corrective osteotomy targeted at the supracondylar femoral area in immature skeletons. Hemiepiphysiodesis might be a more feasible option in those cases as it provides the surgeon with the choice to remove the staples when necessary. Journal of Pediatric Orthopaedics B 2015, 00:000–000 Copyright © 2015 Wolters Kluwer Health, Inc. All rights reserved.

Keywords: bone diseases, dysplasia epiphysealis hemimelica, limb-length discrepancy, skeletal dysplasia, Trevor’s disease, tumor-like conditions

Introduction
Trevor’s disease, also known as dysplasia epiphysealis hemimelica (DEH), is a rare nonhereditary developmental skeletal disorder that affects epiphyses. First reports of this type of dysplastic lesions were presented in 1926 by Mouchet and Berlot [1]; at that time, the disorder was named tarsomegaly.

In 1950, Trevor reported eight cases of DEH and used the term ‘tarso epiphyseal aclasis’. In 1956, Fairbank described 14 additional cases and suggested the term ‘dysplasia epiphysealis hemimelica (DEH)’ [2,3].

Owing to their contributions of this disease, today this disease is called with their names as Trevor’s disease or Trevor–Fairbank disease.

This disease usually affects lower extremity joints, especially the ankles and knees [2,3]. The incidence rate of DEH is estimated to be one per million [4]. No racial predilection is known to exist and DEH is most commonly encountered in males, with a male to female ratio of 3 : 1 [4–6]. The usual symptoms are the appearance of an osseous protuberance on a single side of the knee, ankle, or foot joint, which gradually increases in size [7,8]. The disease is usually restricted to the medial or the lateral side of the limb (hemimelic), but reports covered in the literature studied usually included medial side involvement [9].

Three clinical scenarios are presented:

(1) Localized: affecting the hind foot or the ankle only.
(2) Classic: with a hemimelic distribution of the lower extremity.
(3) Generalized: involving the entire lower extremity from the pelvis to the foot.

Radiological diagnostic criteria of the disease were clearly defined by Fairbank [2] as follows:

(1) Nonuniform bone growth.
(2) Unconnected ossification centers related to epiphyses [2].

DEH is generally classified as bone dysplasia [4].

The main aim of this article is to pinpoint the unique features of Trevor’s disease and to show possible treatment modalities by performing a review of the recent literature on this disorder.

Case 1
A 23-year-old man presented to our clinic with pain in the left ankle and a limp. The pain had been ongoing for 2 months and it began as a sprain to the left ankle sustained while playing football. The pattern of pain changed with movement and sometimes night pain manifested in the anterior region of the ankle.

Physical examination indicated pain and marked tenderness in the anterolateral region of the ankle without any prominent swelling. Varus and valgus stress tests of the ankle proved to be negative, and passive dorsiflexion was limited and painful. The American Orthopedic Foot and Ankle Society (AOFAS) score of the ankle was 28.

Plain radiographs showed an anterior spur-like bony protuberance at the distal end of the tibia (Fig. 1). MRI determined bone marrow edema adjacent to the spur-like bony protuberance and synovial effusion around the lesion; it also confirmed that all the ligamentous structures were normal.

On a computed tomography scan, an anterior spur or residual small bony protuberance at the end of the anterolateral border of the distal tibia was detected (Figs 1 and 2a–d).

A conservative treatment regimen was prescribed, which included rest, ice application, small heel lift exercises, and NSAIDs. This conservative treatment was not successful and the pain reappeared within 3 days.

However, the patient’s symptoms were resolved after an intralesional local anesthetic injection using fluoroscopy. On the basis of these clinical and radiological findings and a positive local injection test, the source of the pain was considered to be an anterior spur.

After the failure of the conservative treatment, surgical intervention (arthroscopic excision) was considered (Table 1).

Under spinal anesthesia and with a tourniquet applied, routine initial diagnostic arthroscopy was performed, which indicated synovial inflammation and a spur-like bony protuberance. Intraoperative physical examination indicated an anterior impingement between the talus and neck and the bony protuberance. Arthroscopic excision was performed (Fig. 2a and b) using a small osteotome. A smooth tibial side was achieved following excision. The results of this examination confirmed that excised material was consistent with an osteochondromatous bony structure.

The pain and the limitation of ankle dorsiflexion disappeared after a week and the AOFAS score increased to 98 within 2 months in the postoperative period. This case is one of our collections, previously published by us as a case report [8].

Case 2
In February 2010, a 13-year-old boy presented to our clinic with a 7-month history of pain and a bony mass (3 × 4 cm) on his right ankle. Pain occurred after playing football or walking a long distance.

Examination indicated a mass that restricted ankle dorsiflexion at the level of the lateral malleolar area. It was of an ossific nature with a lobulated appearance and reached the bones, mimicking synovial chondromatosis in the joint (Fig. 3a–d).

Surgery was indicated as the ankle was swollen and the patient had pain during physical activity. A skin incision over the mass was made under general anesthesia with a tourniquet applied. The anteroinferior syndesmotic ligament was detached. The mass originated from the epiphysis and was connected to the fibula, pushing the posterior side. Osteochondral structures were loosely
connected to each other and the appearance was similar to synovial chondromatosis (Fig. 3b and c). The mass was completely excised from the epiphysis (Fig. 3d). The detached anteroinferior tibiofibular ligament was anatomically sutured (Table 1).

At the ninth month of follow-up, the patient returned to his previous level of activity. The ankle range of motion was normal without pain and disability. This case is one of our collections, published previously by us as a case report [9].

Case 3
In March 2013, a 12-year-old boy presented to our clinic with a 9-month history of pain and a bony mass (3 × 3 cm) located at the right ankle. Pain occurred after walking long distances.

Examination indicated a protuberant mass at the retrocalcaneal area that restricted ankle plantar flexion (Fig. 4a and b). It was ossified with a lobulated appearance and located within the retrocalcaneal area of the ankle joint (Fig. 4a and b).

The patient refused surgery; thus, we adopted a follow-up procedure. No major deformity or apparent walking disability was detected during his last follow-up appointment. We intend to continue following the progress of this patient.

Case 4
In November 2012, a 4-year-old boy was referred to our clinic with limping, deformity, and limb-leg discrepancy (LLD). Physical examination indicated a lobulated bony protuberance at the posteromedial ankle and bony protuberance at the lateral side of knee, with a deformity of the left lower leg (Fig. 5a–d, genu varum).

Plain radiographs of the lower extremity showed an osteochondroma-like bony mass at the posteromedial side of the ankle and physical overgrowth on the lateral epiphysis of the left distal femur. In addition, involvement of the lateral half of femoral head epiphysis was observed (Fig. 5b–g).

After a 19-month follow-up period with close observation (June 2014), three different decisions were made for three different anatomic scenarios (Fig. 5a, c, g). These were as follows:

1. Excision: for ankle involvement (Fig. 6a).
2. Hemiepiphyses: for the lateral physis of the distal femur and proximal tibial lateral physis (Fig. 6b).
<table>
<thead>
<tr>
<th>References</th>
<th>Number of cases</th>
<th>Entire lower extremity involvements</th>
<th>Hip involvements</th>
<th>DFLE involved cases</th>
<th>Asymmetric involved cases</th>
<th>Treated with osteotomy</th>
<th>Cases treated with epiphysodesis</th>
</tr>
</thead>
<tbody>
<tr>
<td>Trevor [3]</td>
<td>8</td>
<td>Case 7. Observation</td>
<td>Case 7. Symetric involvement, treated with valgus heel wedge. No LLD reported</td>
<td>–</td>
<td>–</td>
<td>Case 8. Supracondylar varus osteotomy. Clinical result was not reported</td>
<td>–</td>
</tr>
<tr>
<td>Fairbank [2]</td>
<td>14</td>
<td>Case 3. Femoral head lateral epiphysis + DFLE + PTLE + lateral malleol + talus lateral + symmetric involvement</td>
<td>Case 3. Femoral head lateral epiphysis</td>
<td>–</td>
<td>Case 7. DFLE, lateral malleol, navicular involved. The lesion at lateral malleol was excised</td>
<td>–</td>
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<td>Fairbank [2]</td>
<td>14</td>
<td>–</td>
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<td>–</td>
<td>Case 1. 2-year-old genu valgum, osteotomy details were not reported in the result</td>
<td>–</td>
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<tr>
<td>Smith et al.</td>
<td>6</td>
<td>Case 4. Entire lower extremity</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>Case 4. Entire lower extremity involvement. The osteotomy region was not reported. Immature skeleton</td>
<td>–</td>
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<tr>
<td>Keret et al.</td>
<td>8</td>
<td>–</td>
<td>Case 8. Acetabulum and femoral head whole epiphysis involved. 2 years of age arthroty and acetabular excision at the age 5.5 years, arthroty excision of the mass at femoral head at age 14 years, varus derotational osteotomy (painless limp)</td>
<td>–</td>
<td>–</td>
<td>Case 3. Femoral supracondylar varus osteotomy. Immature skeleton 22 months of age. LLD (1.5 cm longer than normal)</td>
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<td>Keret et al.</td>
<td>8</td>
<td>–</td>
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<td>Case 4. Femoral supracondylar varus osteotomy. Immature skeleton (between 13 months and 3 years), details were not reported in the result</td>
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<td>Keret et al.</td>
<td>8</td>
<td>–</td>
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<td>–</td>
<td>Case 2. Femoral supracondylar varus osteotomy. Immature skeleton 11 months at age 2. Repeat osteotomy. LLD (1 cm shortening)</td>
<td>–</td>
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<tr>
<td>Kettelkamp et al. [5]</td>
<td>15</td>
<td>Case 13. Entire lower extremity. Shoe lift on unaffected side. Details were not reported in the result</td>
<td>–</td>
<td>–</td>
<td>Case 13. Asymmetric involvement (femoral head + DFME + PTLE + distal tibia)</td>
<td>–</td>
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<tr>
<td>Connor et al. [12]</td>
<td>9</td>
<td>Case 5. DFLE, femoral head (lateral epifiz), distal tibia ME, talus entire lower extremity, asymmetric involvement. Treatment: heel rise with observation, 1 cm LLD, 2.5 cm wasting tigh, 1 cm wasting calf</td>
<td>–</td>
<td>Case 5. DFLE, femoral head (lateral epifiz), DTME, talus. Being treated with heel rise with observation. LLD (shortness 1 cm), 2.5 cm wasting tigh, 1 cm wasting calf</td>
<td>–</td>
<td>Case 1. Stapling of distal tibial epiphysis (LLD: 1 cm), she was leading a normal life</td>
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<td>Skaggs et al. [13]</td>
<td>2</td>
<td>–</td>
<td>Case 1. Acetabular involvement. 2 times surgically dislocated with anterior approach. At 8 years follow-up, she was participating in sports with no pain or limp. Case 2. Acetabular involvement. Anterior approach surgically dislocated. At 2-year follow-up, she had full painless ROM of the hip and reported no problems</td>
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<td>Rosero et al. [14]</td>
<td>7</td>
<td>One case had scapular involvement, one case had both distal and proximal femoral head epiphysis involvement, one case had proximal tibial epiphysis involvement, three cases had talus involvement, one case had calcaneus involvement.</td>
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<td>References</td>
<td>Number of cases</td>
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<td>Asymmetric involved cases</td>
<td>Treated with osteotomy</td>
<td>Cases treated with epiphysodesis</td>
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<td>Struijs et al. [15]</td>
<td>7</td>
<td>All cases were at the ankle</td>
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<td>Skriptiz et al. [16]</td>
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<td>Struijs et al. [15]</td>
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<td>Skriptiz et al. [16]</td>
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<td>Wentvreden and Jansen [17]</td>
<td>3</td>
<td>All cases were at the ankle</td>
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<tr>
<td>Fasting and Bjerkeim [18]</td>
<td>4</td>
<td>All cases had genu vagum</td>
<td>–</td>
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<tr>
<td>Bombaci et al. [19]</td>
<td>2</td>
<td>One knee + one wrist</td>
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<tr>
<td>Bakerman et al. [20]</td>
<td>4</td>
<td>All cases were ankle</td>
<td>–</td>
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<tr>
<td>Bhosale et al. [21]</td>
<td>2</td>
<td>All cases were ankle</td>
<td>–</td>
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<tr>
<td>Bosch et al. [22]</td>
<td>9</td>
<td>All cases had immature skeleton. One case had DFME involvement. Excision: one case had wrist involvement, one case had calcaneus involvement, one case had PTE involvement, one case had DTME involvement, 6 cases had talus involvement. 8 patients underwent excision</td>
<td>–</td>
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<td>2nd case had DFLE involvement</td>
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<td>Oberc et al. [23]</td>
<td>6</td>
<td>Details were not reported</td>
<td>–</td>
<td>–</td>
<td>2 hip joint involvement, they did not provide details of the anatomic and follow-up features</td>
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<tr>
<td>Tschauner et al. [24]</td>
<td>1</td>
<td>Lesion at femoral head was excised + pemberton-like</td>
<td>DFLE involved</td>
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<td>Authors</td>
<td>Year</td>
<td>Case Characteristics</td>
<td>Treatment Details</td>
<td>Results</td>
<td>Notes</td>
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<td>Cruz-Conde et al. [25]</td>
<td>2025</td>
<td>Entire lower extremity involved. They operated only hip and performed Smith–Petersen anterior approach, capsulotomy, they surgically dislocated the hip, excised the lesion (last visit: LLD 6 cm)</td>
<td>They performed Pemberton-like acetabuloplasty.</td>
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<td>Mendez et al. [26]</td>
<td>2026</td>
<td>Hip. Entire femoral head epiphysis (acetabulum was not involved), surgically subluxated hip and varus osteotomy. LLD: 2 cm</td>
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<tr>
<td>Linke et al. [27]</td>
<td>2027</td>
<td>Surgical dislocation. Recurrence, 2-year follow-up average results</td>
<td>–</td>
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<td>Wenger and Adamczyk [28]</td>
<td>2028</td>
<td>They performed surgical dislocation, she had no hip complaints and had returned to all activities. Her hip ROM continued to be moderately decreased compared with the uninvolved side</td>
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<td>Haddad et al. [29]</td>
<td>2029</td>
<td>They performed excision of the mass located at femoral head and neck, LLD and joint limitation remains</td>
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<tr>
<td>Sherlock and Benson [30]</td>
<td>2030</td>
<td>Lesion was excised from the femoral neck + posterolateral approach utilized</td>
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<tr>
<td>Philips et al. [31]</td>
<td>2031</td>
<td>–</td>
<td>DFLE involvement (excision)</td>
<td>–</td>
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<tr>
<td>Zhu et al. [32]</td>
<td>2032</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>Treated with medial close wedge osteotomy and MCL repair. The patient was 22 years old (mature skeleton). They achieved normal mechanical axis with normal distal femur alignment.</td>
<td></td>
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<tr>
<td>Wheeldon G and Altiok [33]</td>
<td>2033</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>Authors did not perform hemiepiphysiodesis on the involved side (distal femoral medial epiphysis).</td>
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</tbody>
</table>

DFLE, distal femoral lateral epiphysis; DFME, distal femoral medial epiphysis; DTME, distal tibia medial epiphysis; LLD, limb-length discrepancy; ME, medial epiphysis; ROM, range of motion; MCL, medial collateral ligament.
Follow-up for femoral head epiphysis involvement (Fig. 5g).
A skin incision was made on the posteromedial aspect of the ankle with the neurovascular bundle; the posterior tibialis tendon and the muscle belly were identified and protected (see Fig. 6a). The bony mass was excised in portions.

Subsequently, we performed a diagnostic arthroscopy and noted that the involved cartilage portions of the distal femoral lateral epiphysis (DFLE) adapted the joint shape; therefore, we decided not to perform an intra-articular excision of the lesion and ended the arthroscopic procedure next to the physis line of the distal femur and proximal tibia, identified using an image intensifier and a small incision performed along the fascia. Then, the fascia was incised along with parallel lines and the vastus lateralis was identified and retracted. The lateral aspect of the proximal tibia was also prepared for epiphysiodesis.

The temporary hemiepiphysiodesis procedure with four staples (8 mm U staple) was performed for both lateral proximal tibial and DFLE using the Blount [34] technique.

A long leg splint was used on the left lower extremity to prevent postoperative pain.

The deformity was seemingly resolved and the desired overcorrection (slightly genu valgum) was achieved at his latest follow-up appointment after the sixth month of the operation. Staples will be retained for 14 more months and then removed to allow hemimelic involved epiphysis to compensate valgus in to restore the situation to normal (Fig. 6b–d).

Discussion
The main aim of this study is to increase awareness of Trevor’s disease among Orthopedic Surgeons and to underline some important aspects of treatment of this rare disorder. In this article, we have reported a wide selection of clinical presentations of the disease from easily treatable cases with arthroscopic excision to very difficult cases with deformity correction with temporary hemiepiphysiodesis.

A systematic search of the PubMed database was performed and the following research criteria were applied:

(1) Papers written in English or papers from which we could obtain an English written abstract.
(2) Papers examining Trevor’s disease or DEH.

In total, 104 of 151 papers fulfilled the inclusion criteria. Papers written in languages other than English (19), papers from which we could not obtain full-text articles or abstracts, and other issues (28) (articles published in radiology journals, veterinary journals, pathology journals, rheumatology, and pediatric journals) were excluded. On
the basis of the exclusion criteria, we obtained 28 studies on lesions at the upper extremity, one on spinal lesions, and 155 studies on lesions located in the pelvis and the lower extremity.

Fig. 3

(a) Irregularities and small chondromatous masses on the anterior aspect of the ankle joint (yellow free lines and arrows, respectively). (b) Computed tomography showing irregular bone particles (confluent ossific mass) on the lateral distal tibial epiphysis (arrows). (c) Cartilage caps over the bones can be seen. (d) After excision of the mass, the ankle joint and the distal tibiofibular corner are shown. Reprinted with permission from Gökkuş et al. [9]. Copyright [Assistant Editor of Journal of Orthopaedic Surgery (Hong Kong Academy of Medicine Press)], [location of copyright holder]. All permission requests for this image should be made to the copyright holder.

Fig. 4

(a) The slight swelling on the posteromedial ankle (arrows). (b) Posterior cauliflower bony protuberance (arrows).
In the recent literature, only 17 articles included two or more cases whereas all other articles were limited to only one case (see Table 1) [2,3,5,10–23,35].

After searching for unusual locations and treatment methods of this disorder, we found eight articles on location and two articles on treatment methods.

Unusual locations:

1. Symphysis pubis (1) [5].
2. Intercondylar notch (1) [10].
3. Scapula (3) [14,25,36].
4. Spine (1) [29].
5. Sacroiliac (2) [37,38].

Unusual treatment methods

Braman and Stewart [39] treated their patient with proximal humeral involvement using hemiarthroplasty. DeVine et al.’s [40] patient was the oldest patient found...
in the recent literature with knee involvement and arthroplasty was used for treatment.

(a) Intraoperative photo: note the osteochondromatous lesion (arrows) under the neurovascular bundle. (b) Postoperative (6 months) radiograph shows that desired overcorrection (slightly genu valgum) was achieved. (c) Clinical photo of the child: the desired overcorrection (slightly genu valgum) was achieved. (d) Lateral appearance of the child; note the sagittal balance of the involved side (left) is very near the normal side.
Reasons for deformity and development of limb-length discrepancy in Trevor’s disease
The involvement of the affected epiphysis is hemimelic, meaning that either the medial or the lateral part of the ossified center is affected. Therefore, the epiphysis grows in an asymmetric manner. This characteristic hemimelic pathological growth pattern clinically results in deformities and LLDs [41]. Limb overgrowths can be attributed to the growth of multiple epiphyseal centers or to increased blood flow from surgical stimulation [12,25, 34,42–45]. Limb shortening is often secondary to premature physeal closure or can occur as a surgical complication [12,39,46].

Entire lower extremity involvement
A review of the literature showed that only five articles had previously reported lower extremity involvement in such cases [2,3,5,10,12,25]. We closely examined these articles and realized that entire lower extremity involvements are likely to cause LLD and deformity (genu varum or genu valgum). Most of these cases required surgical treatment (Table 1).

Our fourth case had complete lower extremity involvement. When the patient was admitted, we decided to observe the lesions every 3 months over a period of 16 months.

Hip involvement
The recent literature included 11 articles on hip (acetabulum and femoral head epiphysis) involvement. Only Keret et al. [11], Skaggs et al. [13], Mendez et al. [26], Cruz-Conde et al. [25], Linke et al. [27], Sherlock and Benson [30], and Haddad et al. [29] decided to perform surgery (an arthrotomy, surgical dislocation of the hip, or subluxation of the joint). With the exception of Linke et al.’s [27] patient, all other patients had LLD and limping. Some of them even needed corrective osteotomy of the proximal femur (Table 1).

Other authors including Connor et al. [12], Fairbank [2], Obere et al. [23], and Trevor [3] decided on a strategy of monitoring their patients. With the exception of Trevor [3], all reported LLD in their patients, although in Connor et al.’s [12] case, reported LLD was only 1 cm (Table 1).

Our approach to the case with hip involvement
In our case, we observed that femoral head epiphysis involvement had minor effects on the patient’s clinical outcomes and hip movements and also that the acetabulum was not affected. Therefore, we decided to follow up the patient without surgical intervention at this point by observation of the lesions in trimonthly periods for 16 months. The patient’s last examination in November 2014 showed no disability at the hip.

Rarity of the cases treated with hemiepiphysiodesis
Despite many cases of genu valgum and genu varum found in the literature, cases treated with hemiepiphysiodesis were quite rare [10,12,16].

Our approach to genu varum treatment
Our view is that it is preferable to perform a hemiepiphysiodesis rather than a corrective osteotomy as epiphysiodesis is a procedure in which one can control the timing of removing or keeping the staples in place in accordance with the follow-up measures used. We performed hemiepiphysiodesis and achieved the desired slight overcorrection, monitoring again 3 months after the surgery to evaluate the situation and to determine whether any further interventions are necessary. We believe that the overgrowing will continue after the staples are removed; therefore, the slight valgus angle will be resolved after staple removal. This can be attributed to the mild overgrowth potential of the epiphysis involved.

Using Anderson et al.’s [47] normal femur and tibia growth charts, we calculated that the femur grew about 9 cm from birth to the fourth year of life. We measured our patient’s femora–tibial angle to be 10° valgus on the unaffected side and 4° varus on the affected side. This means that the pathologic epiphysis changed to its normal course toward the varus direction at ≈14°. Without epiphysiodesis, our estimation was 24.2° varus, with a growth ratio of 9–22 cm (22 cm growth expected on 16). We chose this age as skeletal growth slows down after 16. After skeletal maturation, the expected physiologic valgus angle between the femur and the tibia should be 5–7°, meaning that we need to achieve 30.2° valgus in total to compensate for the expected 24.2° adverse varus effect of the pathologic epiphysis after removal from the physe. Considering that it took 6 months for 9° correction with hemiepiphysiodesis, we expect to achieve 30.2° correction in 20 months. Therefore, we aim to follow the patient for an additional 14 months. In other words, our goal is to attain 30.2° valgus angle at the knee and we can remove the staples after this desired angle is achieved.

Still, it should be kept in mind that all these details were obtained using mathematical calculations. We are aware of the fact that the growth rate is not directly in proportion with time. However, these calculations can be used to predict approximate results. To the best of our knowledge, no study has been carried out to further
predict the amount of progress after osteotomy or hemiepiphysiodesis.

**An unusual case with a combined method of treatment in terms of genu varum correction**
Nishiyama et al. [48] reported an interesting case diagnosed in a 5-year-old patient. Despite having DFME involvement, the authors also reported varus deformity (which we believe might have been caused by tibial bowing toward the opposite side) and tibial shortening. They performed corrective osteotomy and limb lengthening with distraction osteogenesis (limb lengthening 6 cm), in addition to a monoplanar external fixator, and there was a gradual improvement in the deformity in this case.

**Distal femoral lateral epiphyseal involvement**
Our fourth case had DFLE involvement. When the patient was admitted, we decided to observe the lesions over trimonthly periods for 16 months. At the end of this observation period, we performed arthroscopy and hemiepiphysiodesis on the knee. The last examination in November 2014 showed slight genu valgum at the knee.

Only six articles in English were found in the literature on DFLE involvement [2,3,5,12,22,31]. After a thorough review of these articles, we found that DFLE involvement was strongly associated with entire lower extremity involvement and the undesirable results included LLD and deformity of the knee (Table 1).

**Asymmetrical hemimelic involvement**
Asymmetrical hemimelic involvement of the epiphysis is also rare. Only three articles with three cases included asymmetric hemimelic involvement [2,5,12]. Of these three articles, two showed DFLE involvement. On examining these studies, we found that asymmetric involvements tended to be associated with DFLE. In our case, four asymmetric aspects also existed with DFLE involvement (Table 1).

**Ankles involvement, conservative treatment regimen, arthroscopic, and open excisions**
Good results using non-surgical treatments were reported in the literature [21,42]. We performed an arthroscopic excision on our first case. In this case, the bony protuberances of two fragments had irregular shapes with an ossific nuleonic appearance. In addition, they were surrounded by a cartilaginous cap, which is considered to be a residue of Trevor’s disease on the MRI sagittal slides (Fig. 2). The shape of bony protuberance was not like a triangular spur. These were the main reasons for our suspicion and the fragments were used for pathological examination.

Tol and Van Dijk [49] described the superficial layer of an osteophyte that contained mesenchymal fibrous cartilage. In this case, the superficial layer of the bony protuberance contained pure hyaline cartilage, that is believed to be caused by osteochondroma (Fig. 3). Also, there was no wedge bone formation dissecting the cartilage, in contrast with osteophyte [50].

In the third case, the patient was advised to undergo surgery, but his parents refused to provide consent for the operation. At the last visit, the patient was leading a normal life, albeit with a minor disability in his ankle. In our fourth case, we performed an ankle excision after 16 months of follow-up.

**Role of arthroscopy in treatment**
After our literature review of ankle treatment procedures, our conclusion was that carrying out excisions is a better choice, mainly because a large number of results yielded a good prognosis in many cases. The number of cases in literature were limited in which the lesion was detected in adult life, as was detailed in our first case. It was a remnant of Trevor’s disease that mimicked an anterior spur of the distal tibia. We would like to highlight the possibility of such occurrences and propose arthroscopy as a suitable treatment method in this instance (referred to in the Case 1 section).

Acquaviva et al. [51] classified the lesions as extra-articular and intra-articular depending on the location of the lesion. In extra-articular localization, simple excision of the mass may be enough to produce favorable results. If the lesion is intra-articular, osteotomy might be needed to correct angular deformity [15,38]. In our experience, in contrast to this idea, if the lesion is intra-articular, especially if it is in the knee joint, the surgeon should perform an arthroscopy and attempt to evaluate the lesion surface that faces the joint. If the joint surface of the lesion is adapted to the curvature, it should be left alone and the hemiepiphysiodesis option should be considered to correct the present deformity. In our fourth case, we assessed the knee using arthroscopy and determined that the articular surface was adapted to the condition.

**Distal femoral osteotomy**
After an exhaustive search of all the available literature, data from patients who underwent distal femoral osteotomy were obtained. We closely examined these cases and realized that this treatment method results in LLD and slightly persistent deformity or recurrence deformity in an immature skeleton most of the time [2,3,10–12]. Only Skr iptiz et al. [16], who combined hemiepiphysiodesis with osteotomy, reported minimal residual deformity. Nishiyama et al. [48] reported good results; however, this patient’s characteristics were different (a 12-year-old girl, at nearly the end of her growth period, on whom they performed osteotomy proximal tibia just below the tibial tubercle). Limb lengthening (6 cm) was also carried out.

In contrast, our fourth case presented with genu varum and was treated temporarily using hemiepiphysiodesis.
(our mentioned case was a 4 year old with open physis). Zhu et al. [32] reported one case of genu valgum on a 22-year-old man who had reached skeletal maturity and his physis was closed. They treated this case with corrective osteotomy (medial close wedge osteotomy) and medial collateral ligament repair (Table 1). The patient was followed up for 2 years and normal mechanical axis was achieved, with normal distal femur alignment (Table 1).

**Conflicts of interest**

There are no conflicts of interest.

**References**


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