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A 28-Year-Old Woman with Down Syndrome, Congenital Heart Disease, and a History of Knee Surgery and Plantar Fasciitis, with Hallux Abducto Valgus (Bunion) and Lapiplasty Three-Dimensional Correction Surgery

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Study Design A
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Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Female, 28-year-old
Final Diagnosis: Congenital heart disease • Down's syndrome • flat foot • hallux valgus
Symptoms: Foot pain • knee joint pain • low back and hip pain, gait changes, proximal muscle weakness
Clinical Procedure: —
Specialty: Podiatry • Surgery

Objective: Congenital defects/diseases

Background: Tarsometatarsal joint (TMJ) arthrodesis is common method used for correcting hallux abductus valgus (HAV). Its popularity has grown due to studies revealing HAV's triplanar deformity with frontal plane rotation. This case report presents a 28-year-old woman with Down syndrome, congenital heart disease, and a history of knee surgery and plantar fasciitis, with severe HAV deformity and flexible valgus flatfoot associated with ligamentous hyperlaxity.

Case Report: Examination revealed severe foot deformities, and radiographic studies confirmed the condition. A surgical intervention was planned, and the patient's cardiologist confirmed she was fit for the procedure. The modified Lapidus technique with frontal plane rotational correction included realigning the metatarsal joint, resecting spurs, osteosynthesis material, and arthrosis in the sinus tarsi. After surgery, the patient underwent a recovery period without support for 8 weeks and received appropriate medical care. Radiographs showed successful alignment, and the patient gradually resumed her daily activities. The patient had an uneventful recovery, and postoperative radiographs showed good alignment in all planes.

Conclusions: The hyperlaxity associated with Down syndrome makes the incidence of HAV more frequent, and TMJ fusion is preferable to correction by osteotomy. The modified Lapidus technique with frontal plane rotational correction could be a good technique to achieve satisfactory correction in patients with severe HAV deformity and flexible valgus flatfoot associated with ligamentous hyperlaxity. TMJ fusion is indicated when severe or recurrent rotational component is observed in X-rays.

Keywords: Arthrodesis • Down Syndrome • Flatfoot • Hallux Valgus
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Background

Hallux abductus valgus (HAV), or bunions, is common in older women and involves a metatarsophalangeal joint deformity causing pain and footwear problems. Initially, non-operative methods like proper footwear, NSAIDs, orthotics, splints, and toe spacers are recommended [1,2]. Surgical referral is advised for patients with persistent pain who have exhausted non-operative options and are suitable for surgery; cosmetic concerns alone are not a reason for surgery. Smoking is discouraged before surgery, and in young patients, surgery should be postponed until skeletal maturity for juvenile bunions [3]. The condition can occur in various populations, including individuals with Down syndrome, who are particularly prone to foot and ankle musculoskeletal abnormalities [4]. Down syndrome, or trisomy 21, is a chromosomal disorder associated with intellectual disability and a higher incidence of congenital malformations [5].

The management of hallux abductus in patients with Down syndrome poses unique challenges due to hypotonia and ligamentous laxity associated with the condition [6]. Traditional surgical approaches may be insufficient in providing long-term correction, and the risk of complications is also heightened [7]. In fact, Down syndrome patients who are increasingly active need faster diagnosis and intervention to prevent severe disability. Surgical procedures in this patient population carry higher risks of complications, such as infections and wound healing problems. Therefore, the search for effective and sustainable corrective techniques tailored specifically to this patient population is of great importance [8].

Lapiplasty is an emerging approach that shows promise using triplanar correction, which addresses the three-dimensional deformity associated with HAV [9]. Special implants are used to fix and maintain this new alignment. By considering all 3 planes of motion, triplanar correction offers the potential for improved functional outcomes and long-term stability across first tarsometatarsal joint (TMJ) arthrodesis [10,11]. The initial indication proposed the correction of severe HAV deformity secondary to metatarsus primus varus. Subsequently, multiple surgical approaches, fixation techniques, and indications for this procedure have been described [12-21], one of these indications being hypermobility of the first radius [22,23]. Some cases of complications associated with this procedure include overshortening, overelevation, pseudarthrosis, and nonunion of the TMJ [24,25]. These complications are attributed to osteosynthesis failure and difficulty adhering to a long and adequate postoperative regimen [21,26-30].

Despite the technical difficulty, the increasing use of this procedure is due to the growing number of studies that consider hallux valgus to be a triplanar deformity, which has a rotational component in the frontal plane [31,32]. Pronation or eversion of the first radius has been consistently described [33-35], with the center of rotation being the first TMJ [36]. Following these findings, Dayton et al published numerous case series of surgical treatment of HAV, applying the modified Lapidus technique with rotational correction in the frontal plane. The triplanar correction decreased the intermetatarsal angle, proximal articular set angle (PASA) and HAV angle, with modification of the tibial sesamoid position without the need for soft tissue or capsular rebalancing of the metatarsophalangeal joint (MPJ) [37,38]. To recognize rotation, radiological assessment of rotational position can be performed using AP and axial views of the sesamoids. Aspects of the AP radiograph that can be used to assess rotational position include the lateral round sign of the first metatarsal head, the position of the tibial sesamoid [24,26,27], PASA angle [23], prominence of the medial eminence [20], and the lateral shaft curvature of the first metatarsal [28].

The aim of this article is to contribute to the growing body of knowledge surrounding the management of HAV in individuals with Down syndrome. by presenting a case study of a patient with Down syndrome who underwent triplanar HAV correction and flexible valgus flatfoot associated with ligamentous hyperlaxity. We hope to help clinicians in making informed decisions regarding surgical interventions for this challenging patient population. The article has been written according to the CARE (CAse REport Statement) guidelines for reporting and developing clinical cases in scientific journals [39].

Case Report

A 28-year-old female patient attended the Clínica Recoletas Paracelso podiatry department (Valladolid) in July 2019. She presented a flexible valgus flatfoot and HAV. Her personal history included Down syndrome, congenital heart disease, and left knee surgery for habitual dislocations. She has no known drug allergies and was not currently receiving any treatment.

The patient reported pain in both feet for several years, having been treated with plantar orthoses, which she did not tolerate or failed to reduce her discomfort. The examination revealed ligamentous laxity and severe deformity of the foot, highlighting the flexible valgus position of the subtalar joint and severe HAV (**Figures 1, 2**).

A physical examination was performed, with the following findings: palpable pulses in the pedial (+++) and posterior tibial (+++) arteries of both lower limbs and good capillary refill, indicating good foot perfusion. Her heart rate was 56 beats per minute (bpm), and her blood pressure was 114/74 mmHg. Dorsal plantar (DP) and lateral loading radiographs were taken,

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Figure 1. Preoperative image: Weight-bearing AP view.

Figure 2. Preoperative image. Weight-bearing front view.

with an oblique projection of both feet. Radiographic studies showed a medialized subtalar axis and a lack of congruency of the first MPJ, possibly due to excessive intermetatarsal angle and signs of severe rotation (**Figure 3**).

The procedure chosen was the placement of an implant in the tarsal sinus together with arthrodesis using the technique described by Lapidus. This technique entails a prolonged postoperative period, during the first 6 weeks without support, and a Walker-type boot for an additional 45 days until a sports shoe can be worn.

Understanding the postoperative period and what it entails, she was referred to her cardiologist to rule out any surgical contraindications, which she confirmed at the time of the

Figure 3. Preoperative X-ray: Weight-bearing AP view.

examination. The first days of September were chosen to schedule the operation on the right foot, as this was the one causing the most pain. A preoperative study was conducted, in which all the values were normal in her blood tests. His vitamin D (25 hydroxycholecalciferol) was 30.93 ng/ml (a reference value below 20 is considered a deficit, and between 30 and 100 are normal values). Her electrocardiogram showed sinus bradycardia at 54 (bpm) without significant alterations. The procedure and possible complications were explained and outlined in the informed consent form signed by the patient (and her guardians).

The surgical intervention was performed under conscious sedation, popliteal block, and hemostasis by surgical tourniquet at ankle level (350 mm/Hg). Antibiotic prophylaxis was performed with cephalosporin (cephalexin 2 g). The following steps were performed sequentially during the surgical intervention:

The surgical procedure commenced with a medial incision and dissection through planes to expose the metatarsal-cuneal joint. Subsequently, disinsertion of the peroneus longus muscle fibers at the base of the first metatarsal (M1) was carried out. A guide wire was then placed at the base of M1 to varus the first metatarsal and correct the deformity, with real-time verification under fluoroscopy.

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Figure 4. Postoperative X-ray: Weight-bearing AP view.

Following this, resection and subsequent drilling of the articular spurs were performed, facilitated by the placement of a bone clamp to reduce the intermetatarsal angle while maintaining rotational correction. For the placement of osteosynthesis material, guide wires were first inserted, followed by the placement of two 3.5 mm diameter and 34 mm length FT screws (Arthrex, Naples, FL, USA).

Regarding suturing, Fiberware 2/0 thread (Arthrex, Naples, FL, USA) was used in the capsular tissue, absorbable suture (Vycryl®; Johnson & Johnson, USA) 4/0 in the subcutaneous tissue, and nylon monofilament (Ethilon®; Johnson & Johnson, USA) 4/0 for skin sutures along with approximation stitches.

At this point, ischemia was addressed. For valgus correction, a lateral incision was made in the tarsal sinus area, followed by the insertion of the guide wire, placement of the distractor,

Figure 5. Postoperative X-ray. Lateral plane.

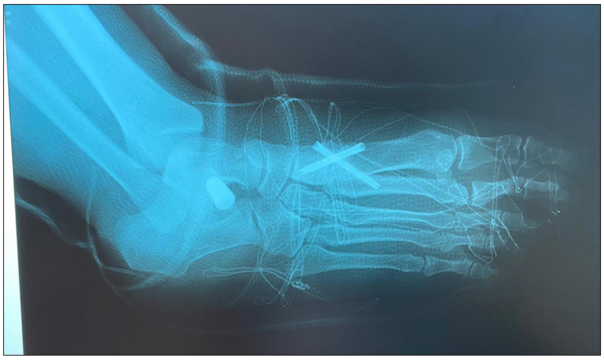

Figure 6. Postoperative X-ray. Oblique plane.

and insertion of the implant while monitoring the direction and ensuring that the length did not exceed the bisector of the talus under fluoroscopy. Finally, postoperative bandaging was performed using a complete splint with a fiberglass bandage (Deltacast®; BSN Medical, Spain)

The postoperative guidelines were home rest, leg elevation, and no weight-bearing for 8 weeks. The prescribed antibiotic treatment was Augmentine 875/125 mg every 8 hours for 5 days. It included Dexketoprofen 25 mg (Enantyum®, Menarini, Spain) every 8 hours, Omeprazole 20 mg every 24 hours, and enoxaparin sodium 4000 IU (40 mg)/0.4 ml (Clexane®, Sanofi, Spain) every 24 hours during the 8 weeks that the patient remained on absolute discharge.

Postoperative radiographs showed good alignment in all planes (**Figures 4-6**). The evolution in the first weeks was positive (**Figures 7, 8**). The suture was removed after 14 days, and weekly dressings were carried out until week 8, followed by follow-ups at weeks 12, 16, and 18, with the next check-up 1 year after the operation. The evolution and dressings were uneventful, postoperative edema was considered normal for the procedure, and the wounds closed satisfactorily. In the second month, 1-month-long support with the Walker boot was

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Figure 7. Postoperative image: Weight-bearing AP view.

Figure 8. Postoperative image: Weight-bearing front view.

allowed. Subsequently, the patient was instructed to walk with a "trekking" boot. At that point, she began progressively returning to daily activities. On 25 June 2020, surgery was performed under the same protocol for the left foot, with no significant differences to report (**Figure 5**).

Discussion

A modified Lapidus technique that incorporates a unique approach to address the limitation in radiographic imaging caused by traditional fixation was introduced. Our modification involves supinating the first metatarsal and securing it with a preferred fixation system, ensuring the desired triplanar correction is maintained. This technique aims to correct not only the lateral deviation of the great toe, but also the axial rotation and transverse plane abnormalities often present in patients with Down syndrome.

First metatarsal rotation has been assessed through different projections. For this study, we used DP, lateral, and oblique radiographic projections under load to obtain the following angulations: first intermetatarsal angle (IMA), hallux abductus angle and position of the sesamoids, assessing the degree of the tibial sesamoid and the angle of sesamoid rotation [40-42]. The results showed a positive correlation between deformity in the transverse hallux plane and in the frontal plane, where the sesamoids are affected together with the angle of inclination of the first radius [40,41]. Other authors, such as Scranton [32] and Mortier [33], used axial imaging of the sesamoids to quantify pronation. The key to the detection of rotation is the edge of the head of the first radius. It will be considered reduced if the medial sesamoid is in the midline of the crest of the first radius, and not reduced if it is in any other position [42]. As for the lateral aspect, the head of the first metatarsal in loading assumes a pronated position, bringing the phalanx into the valgus and increasing its deformity [41,42].

Menz and Munteanu analyzed 95 subjects with HAV using DP radiographs, where they measured angulations that were subsequently correlated with Manchester scale scores. They concluded that the Manchester scale provides a valid proxy for the classification of HAV [43]. A randomized control trial by Welck et al selected a group of 50 subjects with HAV and 50 control subjects, who subsequently underwent computed tomography scanning, and the images were standardized for angle measurement. Using this method, it was possible to assess the position of the sesamoids, the rotation, and the space between the metatarsal and the sesamoid complex. Significant differences were observed between the 2 groups, demonstrating that computed tomography scanning of the foot is a useful method of diagnosing and assessing the degree of sesamoid displacement, as well as evaluating the narrowing of the joint space between the metatarsal and sesamoid associated with sesamoid rotation [44].

Some authors reported that TMJ arthrodesis is indicated in the first X-rays as moderate-to-severe HAV deformities and hypermobility [45,46]. A meta-analysis by Schuh et al [46] compared the corrective power through osteotomies. The corrective power of proximal osteotomies was confirmed with an IMA correction of 8.1°. Although, with a higher complication rate (18.7%), the Chevron osteotomy obtained a higher correction of 8.2°. However, a combination of distal Chevron osteotomy with scarf osteotomy obtained an average correction of 5.3° and 6.2° of IMA, respectively. This would explain the more significant correction obtained after the application of tarsometatarsal arthrodesis [46].

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There is a consistent relationship between the fibular sesamoid and the second metatarsal described in the literature [31]. This observation lends itself to a widely accepted hypothesis of a pathological process in which the first metatarsal deviates from a stable and stationary sesamoid apparatus only in the transverse plane. However, the appearance of the sesamoids on DP radiographs does not reflect the actual subluxation concerning the medial crest and the bisection of the metatarsal shaft [29]. Pronation of the metatarsal alters the way it is seen on the DP radiographic projection, as does an oblique radiograph of the foot [26,27,30]. Dayton et al performed a cadaveric study in which the first TMJ was released and the metatarsal was placed in varying degrees of pronation and supination. With pronation of the metatarsal, the appearance of lateral displacement of the sesamoids was observed on DP radiographs. With supination, the apparent sesamoid displacement was corrected. The metatarsal did not separate from the sesamoid apparatus; instead, rotation altered what was observed on the DP radiographs [26]. Although there is a component of DP sesamoid position that involves the transverse plane, with ridge erosion and actual sesamoid subluxation occurring [19], in general, the appearance of displacement seen on DP radiographs is due to metatarsal pronation.

In fact, the importance of the sesamoids and adjacent soft tissues is shown in this procedure. We consider it essential to reposition the sesamoids under the first metatarsal through a supination rotation of the first metatarsal. For this, we use a joystick guide wire under fluoroscopic control. This indirectly allows realignment of the first toe without operating on the joint capsule, ligaments, or adjacent tendons. Other authors have demonstrated this correction without acting on the first MPJ [37,39].

Regarding whether adequate correction is achieved without touching the MPJ, metatarsus abductus is frequently seen in patients with HAV deformity. The type of procedure should be selected depending on the type of deformity and its degree of severity [45-47]. Lapidus or tarsometatarsal joint fusion is one of the indicated techniques for reducing the IMA and metatarsal abduction (MA) correction [45,46]. In the study by Boffeli et al, 34 patients were included, with an average IMA of 19.4° and postoperative IMA of 9.7°. The mean preoperative Engel angle was 27.4° and was postoperatively reduced by an average of 22.6°. The metatarsus abductus was modified in terms of its degree of flexibility, providing optimal correction in the transverse plane [45,46]. According to Lapidus' theory, hypermobility of the first ray is one of the factors contributing to a valgus position. Correction of this deformity through arthrodesis would be more powerful and long-lasting in this type of deformity [46], being indicated in the first X-ray's as moderate-to-severe HAV deformities and hypermobility [43,44]. In fact, screw fixation shows optimal fixation results, especially if the second screw is inserted from the medial aspect [43]. The screw-plate application offers better stiffness results than the plate-plate application [45].

Menz and Munteanu analyzed 95 subjects with HAV using DP radiographs, where they measured angulations that were subsequently correlated with Manchester scale scores. They concluded that the Manchester scale provides a valid proxy for the classification of HAV [43]. A randomized control trial by Welck et al selected a group of 50 subjects with HAV and 50 control subjects, who subsequently underwent CT scanning, and the images were standardized for angle measurement. Using this method, it was possible to assess the position of the sesamoids, the rotation, and the space between the metatarsal and the sesamoid complex. Significant differences were observed between the 2 groups, demonstrating that CT scanning of the foot is a valid method of diagnosing and assessing the degree of sesamoid displacement, as well as evaluating the narrowing of the joint space between the metatarsal and sesamoid associated with sesamoid rotation [44]. A meta-analysis by Schuh et al compared the corrective power through osteotomies. The corrective power of proximal osteotomies was confirmed with an IMA correction of 8.1°, although, with a higher complication rate (18.7%), the Chevron osteotomy obtained a higher correction of 8.2°. However, a combination of distal Chevron osteotomy with scarf osteotomy obtained an average correction of 5.3° and 6.2° of IMA, respectively. This would explain the more significant correction obtained after the application of tarsometatarsal arthrodesis [46].

In relation to the need for exostectomy and soft tissue techniques, we made the incision over the dorsomedial aspect of the TMJ, exposing it and freeing the joint to allow rotation of the metatarsal to correct the frontal plane deformity. If the correction is found to be in a rigid position, the structures of the lateral complex (hallux abductor muscle) should be released. It is important that the medial aspect of the MPJ capsule should be kept integral to ensure stability and position [48,49]. Once these procedures have been carried out, the positioner and the cutting guide are placed to allow correction in the 3 planes, and the position is subsequently maintained using plates or screws [43,45,48,49]. Therefore, it will only be necessary to intervene in situations in which degenerative changes are observed in the MPJ, both in the capsule and in the dorsal area of the joint [48,49]. If the first metatarsal is corrected from a frontal plane, freeing the structures that prevent rotation, and fixed in the correct position, bunionectomy or other similar techniques will not usually be required [43,44,48,49].

The standard Lapidus procedure, while effective in correcting hallux abductus deformity, can pose challenges in accurately visualizing the achieved correction in postoperative radiographs.

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This limitation arises due to the hardware used in traditional fixation, which can obscure the joint space and bony alignment, hindering precise assessment of the achieved correction. To overcome the radiographic limitations, we propose a modification to the traditional Lapidus technique. In our approach, after performing the necessary osteotomies and realignments, we intentionally rotate the first metatarsal in a supinated position to achieve the desired correction. The rotation allows for a clear visualization of the first tarsometatarsal joint and adjacent structures in radiographs. This procedure is like that carried out in a retrospective case-control study by Laleveé et al in which they showed the importance of supination to reduce pronation of the first metatarsal [48].

Conclusions

The modified Lapidus technique offers a promising solution to the radiographic challenges associated with traditional fixation methods. When there is a rotational component with a severe or recurrent deformity, one indicated technique is TMJ fusion. By emphasizing the importance of triplanar correction and proposing a unique supination approach, we aim to provide surgeons with a valuable tool for achieving optimal outcomes in HAV correction.

In patients with Down syndrome, a complete anamnesis is necessary, followed by a clinical examination, in which we assess the hypermobility of the first radius, the position of the foot in load, and the length of the second toe concerning the first toe and pulses. It is essential for the surgical community to explore innovative modifications that enhance visualization while preserving the integrity of the correction. We encourage further research and clinical validation of this modified technique to improve patient care and enhance the understanding of HAV correction procedures.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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