Foot and Ankle Surgery xxx (2016) xxx–xxx

Contents lists available at ScienceDirect

Foot and Ankle Surgery

journal homepage: www.elsevier.com/locate/fas

Case report

Long term follow-up and development of foot complaints in a surgically treated mirror foot—A case report and review of literature

S.A. Lalé, BSc[a], E.B. Burger, MD[a,*], J.H.J.M. Bessems, MD[b], V. Pollet, MD[b], C.A. van Nieuwenhoven, MD Phd[a]

[a] Department of Plastic, Reconstructive and Hand Surgery, Erasmus MC-Sophia, University Medical Center Rotterdam, the Netherlands
[b] Department of Children's Orthopaedics, Erasmus MC-Sophia, University Medical Center Rotterdam, the Netherlands

ARTICLE INFO

Article history:
Received 24 November 2016
Received in revised form 12 February 2017
Accepted 14 February 2017
Available online xxx

Keywords:
Mirror foot
Long term outcome
Tibia deficiency
Talus deformity
Tarsal abnormalities

ABSTRACT

Background: Mirror foot is a rare anomaly and limited long term follow-up information is available.
Methods: Seven years after operation a mirror foot patient returned with foot complaints and was evaluated using radiographs and clinical examination. A systematic literature search was conducted to study foot complaints in mirror feet.
Results: Different origins of foot pain were considered in our patient; tibia length difference, deformed talus and accessory osseous structures in the tarsal region. Literature search resulted in 118 mirror feet. Based on cases reporting osseous structures, 74.2% showed tibia abnormalities and 94.5% an abnormal tarsal region. Only three cases mentioned a normal talus. Nine cases reported a follow-up period of more than five years.
Conclusion: Osseous abnormalities are not always visible at birth, but are often present. Therefore, detailed examination of the affected limb in mirror foot patients with foot pain is important, in order to localize the origin.

1. Introduction

Polydactyly of the foot is a limb malformation characterized by the presence of supernumerary digits [1]. The formation of fingers and toes during embryogenesis is controlled by sonic hedgehog (Shh). The number of digits and digit identity is influenced by Shh signalling patterns, which is regulated by the Gli3 protein and could cause preaxial polydactyly [2].

Mirror foot, a rare type of preaxial polydactyly, is characterized by mirror-image duplication around a midline axis with a recognizable hallux in the centre [3]. According to several authors, the definition of a mirror foot varies [4,5]. Literature shows an important variability in patterns [5]. Therefore, it is difficult to define the term mirror foot. According to Sudesh et al. less than thirty mirror foot cases have been reported in literature [6]. This number emphasizes the rarity of mirror foot cases in the general population, but exact prevalence is unknown.

Treatment consists of excision of the extra rays to allow fitting of shoes. Despite a lot of case reports about treatment strategies and outcomes, long term follow-up data is often lacking. Little information is present about the foot problems that might occur later in life. In this case report, we present a patient who was operated on a right-sided mirror foot. Seven years after operation, the patient returned with right-sided foot complaints. The aim of this case report is to give more insights in different causes of foot complaints on the long term in patients with surgically treated mirror foot. Furthermore, we performed a literature search in PubMed and Google Scholar for articles written in English or Dutch (Appendix A), to evaluate the presence of these problems in other case reports to reflect on our results.

2. Case report

A boy, born without complications after a full-term pregnancy, was diagnosed with a right-sided mirror foot at birth. No other

* Corresponding author at: Department of Plastic, Reconstructive and Hand Surgery, Erasmus MC, University Medical Center Rotterdam, Room Ee15.91a, Postbox 2040, 3000 CA Rotterdam, Netherlands.
E-mail address: e.burger@erasmusmc.nl (E.B. Burger).

http://dx.doi.org/10.1016/j.fas.2017.02.017
1268-7731/

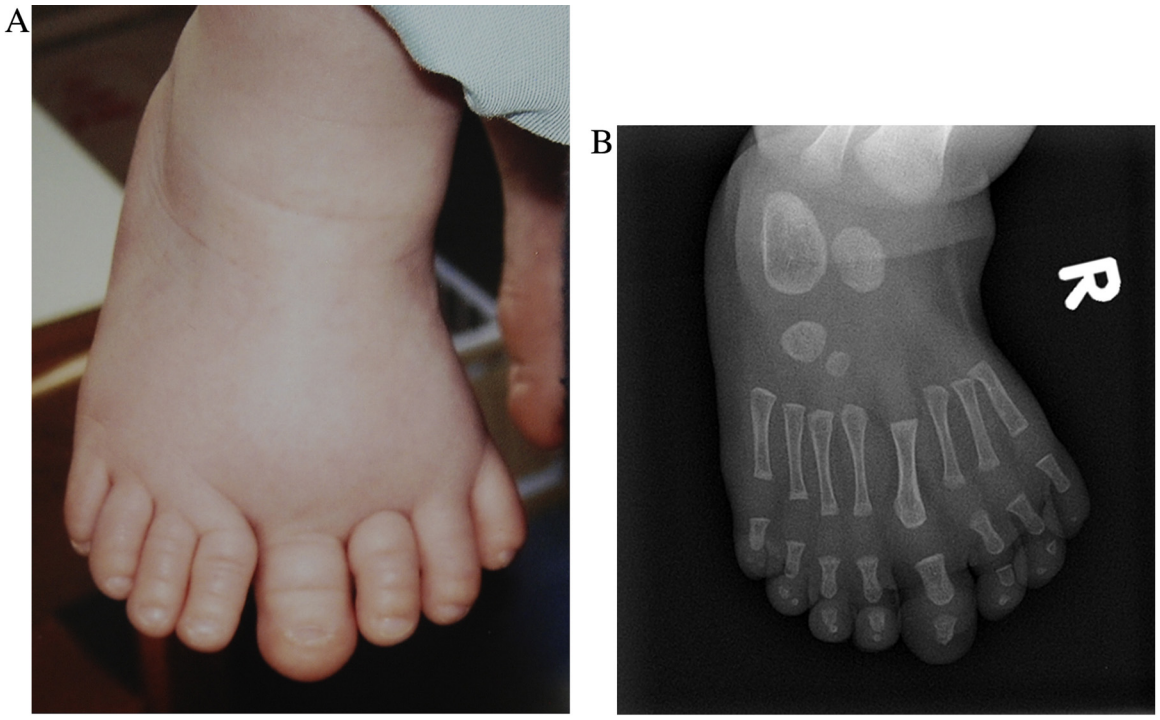

Fig. 1. Preoperative view (a) and radiograph (b) of the right foot.

congenital abnormalities were present. The family history was negative for congenital anomalies. Examination of the right limb showed a mirror foot, with eight toes in total, including an identifiable hallux in the middle flanked by three toes on the medial site of the hallux and four normally developed toes on the lateral side of the hallux (Fig. 1a). Radiographs of the right foot revealed eight metatarsal bones and their associated phalanges (Fig. 1b). Furthermore, radiographs of both lower legs showed a tibial length difference of 0.9 cm in favour of the left leg (Fig. 2). The left lower limb and foot showed no abnormalities. Excision of the three medial rays and reconstruction of the right foot was performed at the age of 11 months. Perioperative an additional extra cartilage tarsal structure was observed connected to the navicular bone and the caput tali. This structure was removed through the cartilage connection. No complications were reported.

At the age of eight years old, the patient returned to the outpatient clinic with pain complaints of his right foot, especially during activities such as running. The pain was located at theanterior part of his ankle. In addition, a mild not painful luxation of the proximal fibular head was present. Furthermore, a difference in the range of motion in dorsiflexion of 10° was observed (right 10°; left 20°). Radiographs of the right foot showed accessory osseous structures (Fig. 3a) as well as a deformity of the talus (Fig. 3b). We decided to treat the patient conservatively, with revision at the out-patient clinic within nine months. After nine months the patient returned for a limb length measurement (photo not shown), showing a 0.5 cm shorter femur on the left side and a 1.0 cm short lower leg on the right side.

The Ethics Committee at the Erasmus Medical Centre Rotterdam, the Netherlands provided approval for the study (MEC-2015-679). Informed consent from parents was obtained.

3. Discussion

Here we present a case of an operated right-sided mirror foot associated with pain complaints of the right leg and foot and impaired ankle dorsiflexion seven years after operation. After evaluation at the outpatient clinic, three different origins of pain can be proposed; presence of accessory osseous structures, dysplasia of the talus, and difference in length between the two legs. To study different causes for foot complaints in mirror feet, we conducted a systematic literature review.

During our search, we noticed that the definition of mirror foot is diverse, due to the variability in patterns. Also different terminologies are used to describe this specific type of foot duplication, the most common terms are diplopodia and mirror (-image) polydactyly. The precise definition of these terms depends on the author. We excluded cases not diagnosed with these terms, even though these cases showed similarities. In total 78 patients, with 118 mirror feet, were identified. All patients were sorted based on the type of tibial deficiency, talus abnormalities, abnormalities in the tarsal region and follow-up time.

In our patient we observed a minimal tibial length difference at birth. To investigate if this is common in mirror foot patients, we searched cases that showed tibial deficiencies and sorted them on type of deficiency (Fig. 4). We identified 11 patients (13 feet) [4,7–13] with tibial hypoplasia, ranging from mild to severe, in combination with a mirror foot. A precise measurement of the tibial length was not always available and therefore these cases were difficult to compare with ours. Four case reports have measured that the affected limb was shorter compared to the unaffected limb. The length differences varied from 3.0 cm to 23.0 cm [7–9]. In 97 feet the tibial development was evaluated and in 74.2% of the cases it was abnormal. Information concerning pain symptoms in these patients was not available. One case report [14], showed a patient diagnosed with polydactyly and normal tibial development at birth. However, hypoplasia of both tibias was observed later in life [14]. This could suggest that tibial length difference is not always detectable at birth and could develop over time or that it was missed due to the minimal difference. Unfortunately, long term follow-up is lacking in this studies. Although our patient did show a tibial length difference, when comparing the entire limb, there is only a difference of 0.5 cm in

Please cite this article in press as: S.A. Lalé, et al., Long term follow-up and development of foot complaints in a surgically treated mirror foot—A case report and review of literature, Foot Ankle Surg (2017), http://dx.doi.org/10.1016/j.fas.2017.02.017