Possible mistakes in prediction of bone maturation in fibular hemimelia by Moseley chart

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Abstract The purpose of this study was to establish a nomogram in order to predict limb length discrepancies in children with unilateral fibular hemimelia more accurately. In 31 children with unilateral fibular hemimelia the femoral-tibial length and skeletal age were determined an average of seven times per case by sequential radiographs during growth. From the data, a skeletal age nomogram was developed which shows a steeply declining mean skeletal age pattern in unilateral fibular hemimelia (the slope in girls was -0.59 and in boys -0.64). This nomogram crosses the normal mean skeletal age line of the Moseley straight-line graph at 10.5 years in girls and at 12 years in boys, and continues to decline until maturity. The results demonstrate an abnormal skeletal maturation process in patients with unilateral fibular hemimelia. The consistently declining steep skeletal age nomogram in unilateral fibular hemimelia makes prediction of skeletal maturity and limb length discrepancy inaccurate by the standard predictive methods particularly when using early skeletal ages. The skeletal age nomogram from our data determines skeletal maturation in children with unilateral fibular hemimelia more accurately, and allows a correct prediction of limb length discrepancy.

Introduction

Children with unilateral fibular hemimelia can have severe leg length discrepancy and the treatment requires adequate

growth prediction of the lower extremity. Contralateral epiphyseodesis and ipsilateral limb lengthening are common procedures used to equalise limb length in patients with fibular hemimelia. Epiphyseodesis of the contralateral distal femoral or proximal tibial growth plates has been used since Phemister described the technique in 1933 [1–11]. This relatively simple operation is an excellent method to correct a mild limb length discrepancy if the growth of the limbs can be accurately predicted. Limb lengthening is a more difficult procedure to correct more severe discrepancies; however, it also requires accurate limb length discrepancy prediction [12, 13]. If growth cannot be accurately predicted, over or undercorrection of the limb length may result at maturity [1, 5, 7, 9]. Difficulties in predicting the growth include variability of skeletal age estimation [3–5, 12, 14], unusual skeletal maturation patterns in some diseases [1, 5, 8, 15, 16], and inaccurate calculations by the physician [1, 5, 8].

In patients with unilateral fibular hemimelia, we observed an abnormal pattern of skeletal age maturation, which made prediction of growth inaccurate. The purpose of this paper was to evaluate sequential skeletal ages of patients with unilateral fibular hemimelia to determine the pattern of maturation and to create a nomogram which provides accurate prediction of skeletal age maturity.

Materials and methods

Thirty-one children who had congenital unilateral fibular hemimelia were followed up from youth and had more than four sequential radiographs taken at least one year apart to determine bone age and limb length, and were entirely treated by the senior authors in the Alfred I duPont Institute. Sixteen patients were female and 15 were male. In 16 patients the unilateral fibular hemimelia occurred on

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the left side, 15 on the right. The average age of the patients at the first visit with limb length and skeletal age estimation was five years and four months (range, three years and four months to six years and eight months). The average age at final follow-up was 19 years and five months (range, nine years and four months to 27 years and two months). Twenty-six of 31 patients with unilateral fibular hemimelia were followed up until skeletal maturity. Five patients were still immature at the time of this evaluation and their average age was 11 years and eight months (range, nine years and four months to 13 years and two months).

The leg length discrepancy and bone age were determined during growth an average of 6.8 times per patient. The orthoroentgenograms were performed by the standard technique [17]. A radiograph centimeter scale was placed beneath the limbs to provide measurements directly on the orthoroentgenogram, along the femur and tibia. Antero-posterior radiographs were made with the tube centred over the hip, knee and ankle joints. The skeletal age was determined by comparing the antero-posterior radiograph of the left hand-wrist with the skeletal developmental atlas of Greulich and Pyle [18]. All of the skeletal ages were determined by consensus of a paediatric radiologist and two of the authors. The data of each patient was plotted to the nearest skeletal age line of the Moseley straight-line graph [19]. The boys and girls skeletal ages were plotted (average 9.2 on the skeletal age line) to create a skeletal nomogram with mean and one standard deviation (Fig. 1). To obtain the best fit line as skeletal age nomogram for patients with unilateral fibular hemimelia, we used the linear regression analysis method.

Results

From our data of skeletal ages of children with unilateral fibular hemimelia, the mean skeletal age with one standard deviation was plotted on the Moseley straight-line graph to create a nomogram line (Fig. 1). Our skeletal age data of children with unilateral fibular hemimelia does not follow the normal maturation pattern. The mean skeletal age of these patients between the ages of four to six is delayed when compared to normal development. With growth (between the ages of six to maturity), the mean skeletal age of the patients with fibular hemimelia declines, which demonstrates a more rapid pattern of maturation than in normal development. This declining mean skeletal age line crosses the mean normal age line of the Moseley straight-line graph at 10.5 years in girls and at 12 years in boys (Fig. 1). After the “cross points” the mean skeletal age line of our patients showed a further decline, and the patients with unilateral fibular hemimelia reach skeletal maturity before normal children (Fig. 1). Both girls and boys with unilateral fibular hemimelia show a similar declining

Fig. 1 Skeletal age nomograms of girls and boys with unilateral fibular hemimelia, which was created by plotting the mean skeletal ages with one standard deviation on the Moseley straight-line graph

pattern of maturation, and the slope of the declining mean skeletal age nomogram line is -0.59 (r squared= 0.92) in girls and -0.64 (r squared= 0.88) in boys (Fig. 1). The angle between the horizontal line in the Moseley straight-line graph and the newly created declining mean skeletal age line of the patients with unilateral fibular hemimelia is 31 degree in girls and 33 degree in boys.

The abnormal pattern of growth in children with unilateral fibular hemimelia causes an error in predicting limb length discrepancy at maturity if the Moseley straight-line graph is used without the “corrective” nomogram. Examples of girls and boys with unilateral fibular hemimelia with 20 percent growth inhibition of the shorter leg are given to demonstrate the error. If maturity in girls with unilateral fibular hemimelia is determined from the early mean skeletal ages of four to five years by a line parallel to the mean line of the Moseley straight-line graph, the length of the normal leg would be 88 cm. When maturity is determined from the later mean skeletal ages of 12 to 13 years, the length of the normal leg would be 75 cm (Fig. 2). When the same is determined for the boys, the normal leg length would be 98 cm and 80 cm, respectively. This represents a large error in predicting the length of the normal leg at maturity.

If a girl with unilateral fibula hemimelia has a 20% inhibition of growth in the shorter leg and maturity in the girls is determined from the early mean skeletal ages of four to five years by a line parallel to the mean line of the Moseley straight-line graph, the length of the “short” leg would be 70 cm. When maturity is determined from the later mean skeletal ages of 12 to 13 years, the length would be 60 cm (Fig. 2). When the same is calculated for boys the length of the normal leg would be 78.5 and 64 cm, respectively. This represents an error in determining the length of the “short” leg at maturity. When the predicted discrepancy is determined from skeletal ages there is a mean 3-cm error in girls and a mean 3.5-cm error in boys.

Discussion

There are several methods to predict growth and limb length discrepancy in children [4, 19–21], and the accuracy of these methods is debatable. The most commonly used method to predict limb length discrepancy and determine maturation is the Moseley straight-line graph method [1, 3, 5, 7, 10, 12, 14, 19, 22], which is a derivation of the data published by Anderson et al. [16, 21, 22], and which is easy to visualise because of the graphic format [14]. The accuracy of predicting limb length discrepancy by the Moseley straight-line graph is supported by only a small number of outcome studies [1, 7–10, 22]. Unfortunately,

Fig. 2 If maturity in girls is determined from early mean skeletal age the length of the normal leg would be 88 cm, and when maturity is determined from late mean skeletal age it would be 75 cm. If maturity in girls is determined from early mean skeletal age the length of the shorter leg (with 20% growth inhibition) would be 70 cm, and when maturity is determined from late mean skeletal age it would be 60 cm

these papers have patients with a variety of different aetiologies, some being congenital deformities and others being acquired conditions [3, 5, 10, 14, 19, 22]. The accuracy of this method is less reliable under the age of ten years [23] and it varies because skeletal age determination is difficult and because many cases do not have a linear pattern of growth [12–15, 17, 24, 25].

According to the results of Timperlake et al., 17 patients from a group of 35 achieved an acceptable limb length discrepancy at maturity (limb length discrepancy less than 1.5 cm) after epiphyseodesis times using the Moseley straight-line graph [9]. A good result was achieved in 90% (limb length discrepancy less than 0.7 cm) of the patients treated by epiphyseodesis in the series of Poratz et al. [7]. Inan et al. found that the Moseley straight-line method accurately predicted the timing for percutaneous epiphyseodesis in all of their 97 patients but one who had hemihypertrophy [10]. Lampe et al. found that accuracy of the Moseley straight-line graph can be limited by a variable pattern of skeletal maturation in some patients [15, 17, 24]. They emphasised the importance of a sufficient number of preoperative measurements to develop an accurate graph. For this reason, they suggest the child be referred to a “limb length clinic” at an early age. In the series of Blair et al., there were 45 failed epiphyseodeses, and in ten the cause of failure was an inadequate operative epiphyseodesis. In the remaining 35 cases, the causes of failure were secondary to errors in estimating the proper skeletal age for operation due to inaccurate or incorrect use of Green and Anderson growth prediction tables [1]. Stephens et al. found that the Green-Anderson charts and the straight-line graph of Moseley both provide an accurate mean of predicting future growth and determination of skeletal age of epiphyseodesis [8]. Aguilar et al. found that the Moseley method is as accurate as the multiplier method in predicting bone maturity lengths but less accurate in predicting limb length discrepancies at maturity after epiphyseodesis [26, 27]. Dewaele and Fabry were not able to significantly improve their results in timing the skeletal age for epiphyseodesis by using the Moseley straight-line graph, and their principal source of error was in estimating the bone age [3]. Kelly et al. emphasised that determination of puberty is essential to reduce errors in timing of epiphyseodesis [28]. Little et al. found that the Gruelich and Pyle skeletal age data could not be shown to increase their accuracy in predicting outcome over serial chronological data, and thus its value in predicting limb length inequality is thought to be limited, regardless of the method used, and unpredictable results occur in a proportion of patients [14]. Evaluation of these papers allows us to conclude that skeletal age determination and variation in normal growth patterns affect the ability to predict growth of the longer (normal) limb. In the abnormal (shorter) limb

additional factors such as disease process and certain treatments may affect the predictability of the growth. For example, Sharma et al. reported severe and unpredictable growth retardation of the tibia after limb lengthening [29].

Unilateral fibular hemimelia is thought to be one of the most predictable diseases for determining limb length discrepancy. Shapiro showed that hemiatrophy (anisomelia) presents a type I growth pattern, which is linear in the Moseley straight-line method [15]. Our data shows patients with unilateral fibular hemimelia have a different pattern of skeletal maturation from normal children. If children with fibular hemimelia are plotted on the Moseley straight-line graph as if they were normal children, the prediction of limb length discrepancy at maturity will be inaccurate. However, if they are plotted using our newly formed reference nomogram line to create the best fit parallel, the prediction will be much more accurate. In this study we had a large series of patients with unilateral fibular hemimelia who had sufficient skeletal age data to evaluate the pattern of maturation. In our group of patients between the ages of four and six the mean skeletal age nomogram line shows immaturity when compared to the normal skeletal age nomogram (Fig. 1). Between the ages of six to maturity, the skeletal age nomogram declines to below the mean age line of the Moseley straight-line graph (Fig. 1). This demonstrates an acceleration of the skeletal maturation process. The mean skeletal age nomogram shows the same declining character in both girls and boys after six years of age (Fig. 1). This declining skeletal age line crosses the mean age line of the Moseley straight-line graph at 10.5 years in girls and 12 years in boys, and continues to decline until maturity. This makes us wonder if unilateral fibular hemimelia involves a general skeletal maturation process rather than being limited to just one limb.

The strengths of this paper are that this series consists of a relatively large group of children with unilateral fibular hemimelia, the children were followed up for many years, and multiple skeletal ages and orthoroentgenograms were obtained yearly. The weakness of the paper is that we were not able to determine the ultimate natural history of limb growth in these children with unilateral fibular hemimelia. Ethically, we could not follow them to maturity without treatment of such a severe deformity.

In conclusion, younger children with unilateral fibular hemimelia have an abnormally immature skeletal age, and as they grow their skeletal age reaches maturity more rapidly than normal. The skeletal age nomogram of patients with unilateral fibular hemimelia consistently declines when compared to the normal skeletal age graph. If the Moseley straight-line graph or the Green-Anderson charts are used for children with unilateral fibular hemimelia, the prediction of limb length at maturity will be inaccurate because they have a different pattern of skeletal maturity

from normal children. The skeletal age nomogram from our data allows a more accurate prediction of skeletal maturation in children with unilateral fibular hemimelia.

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