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Clinical and radiological characteristics of forearm deformities in children with multiple hereditary exostoses

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Introduction The incidence of forearm deformities in children with multiple hereditary exostoses (MHE) ranges from 30 to 80 %. There are few studies of deformities of the forearm in MHE patients in the literature that describe not only the location of exostoses and position of the head of the radius but also the true variants of forearm deformities. The **aim** of the study was to investigate forearm bone deformities in patients with multiple hereditary exostoses. **Materials and methods** Radiographs of the bones of the forearm in 84 patients (151 limbs) diagnosed with multiple hereditary exostoses in the age of four to 17 years who were treated at our institute from 2004 to 2018 were retrospectively analysed. The study involved 47 boys and 37 girls; 67 patients (80 %) had bilateral lesions, and 17 patients (20 %) had lesions of only one upper limb. Patients were divided into four groups depending on the type according to the Jo& Jung's classification. The deformities were evaluated based on radiological methods in accordance with the reference lines and angles for the forearm bones. **Results** The most common variants of forearm deformities were revealed: varus recurvatum at the border of the upper and middle third of the ulna (55 %), varus recurvatum at the border of the upper and middle third of the ulna associated with varus of the radius in the middle third (15 % of cases); as well as their combinations accompanied by dislocation or subluxation of the radial head (30 %). RAA (radial articular angle) and RB (radial bowing) did not have significant difference in various types of deformities of the forearm according to Jo&Jung's classification. **Conclusion** The study of the variety of forearm deformities in children due to multiple hereditary exostoses will assist in a differentiated approach to the choice of surgical treatment methods depending on the type of deformity.
Keywords: multiple hereditary exostoses, forearm deformities, classification

INTRODUCTION

Multiple hereditary osteochondromas (HMO) also known as multiple hereditary exostoses (HME) is a genetic autosomal dominant inherited disorder of the human skeleton; primary spontaneous mutations are observed in 20 to 30 % of cases. Its incidence is 1:50,000 newborns. The pathology may manifest itself already in infants of the first year of life.

Multiple osteochondromas is a frequent pathology in the pediatric practice. Its incidences make from 16 to 43 % of all cases of tumours, tumour-like and dysplastic bone diseases, as reported. Deformities of forearm develop due to multiple exostoses in 30 to 80 % of cases: ulnar (83 %) and radial (17) club hand among them [1, 2, 3]. Limb length discrepancy, angular deformities, and decrease in the range of motion in the adjacent joints as well as pain due to local irritation of muscles, tendons and nerves due to exostosis are observed. Progression of angulation may result in instability in the elbow and wrist joints.

Three classifications that reflect deformities of the forearm bones due to HMO were found in the literature sources on this topic.

The classification developed by A.P. Pozdeev and L.Yu. Khodzhaeva is based on the ulnar deviation of the hand, restriction of movements in the adjacent joints, presence/absence of deformities of the bones of the forearm, magnitude of the forearm shortening, presence/absence of rotation restriction, decentering /subluxation /dislocation of the radial head. Thus, they distinguish five grades of ulnar club hand [3]:

Grade I: ulna deviation within 10–15°. The hand acquires its central position actively or passively, full range of motion in the adjacent joints;

Grade II: ulna deviation up to 30°. The hand acquires its central position actively or passively; there is arch-like deformity of the forearm bones and its shortening up to 2.5 cm; restricted rotation due to decentering and radial head subluxation;

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Grade III: ulna deviation is more than 30°. The hand does not acquire the centered position; forearm bones deformity and its shortening up to 3 cm, sharp restriction of rotation due to subluxation and full dislocation of the radial head;

Grade IV: ulnar deviation more than 30°; the hand does not acquire the centered position; full dislocation of the radial head; deformities of both forearm bones; rotation within 10–15°;

Grade V: ulnar deviation not more than 30°, lesions of distal parts in both bones; dislocation of the redial head; deformity of the metadiaphyses of both bones; considerable shortening of the segment, muscle hypotrophy, flexion contracture of fingers.

The second classification was developed by Masada and Ono, and evaluates the relationship of osteocartilaginous exostoses location and presence/absence of radial head dislocation (Fig. 1) [4, 5]:

Type I: osteocartilaginous exostoses are located in the distal ulna and there is no radial head dislocation;

Type IIA: radial head dislocation and osteocartilaginous exostoses in the proximal methaphysis of the radius, shortening of the ulna;

Type IIB: osteocartilaginous exostoses are located in the distal ulna associated with radial head dislocation, no ulna shortening;

Type III: osteocartilaginous exostoses are located in the distal radius, without radial head dislocation, shortening of the radius.

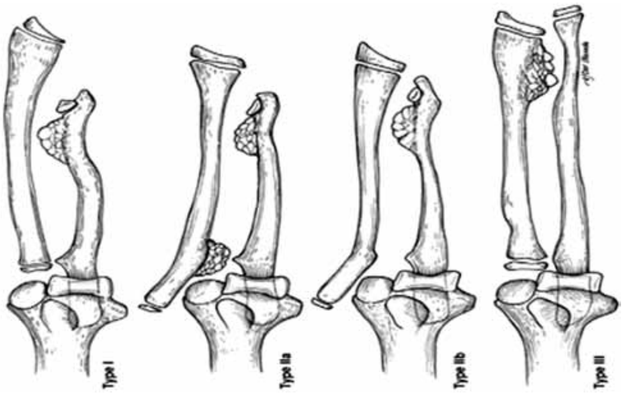

Fig. 1 Schematic presentation of anatomical abnormalities in accordance with the classification of Masada K., Ono K. (1989)

A.R. Jo et al. retrospectively analysed the radiographs of 53 individuals with HMO (102 upper limbs) and supplemented the latter classification with two more types of deformities:

Type IV: osteocartilaginous exostoses located in the distal parts of both bones

Type IVA: osteocartilaginous exostoses located in the distal parts of both forearm bones combined with radial head dislocation;

Type IVB: osteocartilaginous exostoses located in the distal parts of both forearm bones without radial head dislocation (Fig. 2) [7].

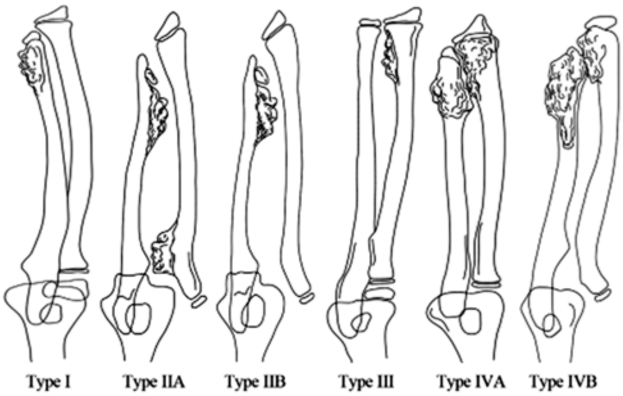

Fig. 2 Schematic presentation of anatomical abnormalities in accordance with the classification of Jo A.R., Jung S.T. (2017)

However, these classifications do not fully disclose the entire variety of deformities of the forearm bones in children due to osteocartilaginous exostoses, but indicate only the location of exostoses and the position of the head of the radius.

Thus, a combination of radial head dislocation with multiplanar deformities of the forearm bones leads to limitation of rotation and loss of function of the upper extremities [7–13]. Peterson suggested that preventing the development of forearm bone deformities in patients with MHO, in particular radial head dislocation, is the most important goal of early surgical treatment [14].

The reference values of the long bone angles, proposed by Solomin L.N., have found application in the work of practicing orthopedists dealing with correction of deformities [15]. Most frequently, these angle values are used in the treatment of lower limb deformities. A few works has been devoted to correction of forearm deformities. There are scarce literature sources that describe deformities of the forearm bones in MHO patients not only from the point of location of exostoses and position of the head of the radius, but reveal true variants of deformities [16, 17].

Understanding the development of deformities and their types, risk factors associated with the possibility of dislocation of the radial head will assist in identification of patients at risk who need early interventions to prevent functional disorders of the upper limb [18, 19].

The aim of the work was to study the deformities of the forearm bones in patients with multiple hereditary osteochondromas.

MATERIAL AND METHODS

Radiographs of the forearm bones in 84 patients (151 limbs) diagnosed with multiple hereditary exostoses in the age of four to 17 years who were treated at our institute from 2004 to 2018 were retrospectively analysed. Sixty-seven patients (80 %) had bilateral lesions, and 17 patients (20 %) had lesions of only one upper limb. Patients were examined at preoperative planning; informed consent was obtained from patients or their legal representatives about the inclusion of their data in the study.

Patients were divided into groups according to Jo& Jung's classification (modified classification of Masada). An orthopedic examination assessed the range of movements (supination / pronation); X-rays and computed tomography of the bones of both forearms were taken in two projections (anteroposterior, lateral). The deformities in the forearm bones were evaluated in accordance with the reference lines and angles: the proximal radial neck-shaft angle (PrNSA), the distal epidiphyseal angle of the radial bone (DEDA), "anatomical axis bowing" in every third for the forearm bones in the sagittal and frontal planes. The angle of inclination of the articular surface of the radius (RAA, radial articular angle), the angle of bowing of the radius (RB, radial bowing), and the shortening of the ulna with respect to the radius were also determined.

In accordance with the Jo&Jung classification, the patients were divided into the following groups (comment: the number was calculated relative to all limbs (151), since one and the same patient could have Type 1 deformity in one limb and Type IV deformity in the other):

Type I (osteocartilaginous exostoses are located only in the distal ulna and there is no radial head dislocation) was detected in 37 patients (24 % of cases).

Type III (osteocartilaginous exostoses are located in the distal radius and there is no radial head dislocation or radial shortening) was identified in 27 patients (18 %).

Type IVA group (osteocartilaginous exostoses are located in the distal parts of both bones combined with radial dislocation) was most numerous,71 patient (47 %).

Type IVB (osteocartilaginous exostoses are located in the distal parts of both bones without radial head dislocation) was detected in 13 patients (9 %).

Club hand of varying severity was present in 100 % of cases. We revealed a significant preservation of both supination and pronation in patients with Type III of this pathology. A significant limitation of both supination and pronation was detected in patients with exostoses located in the distal part of both bones of the forearm. In this case, the dislocation of the radial head did not significantly affect the range of joint motion. The data obtained are presented in Table 1.

According to the findings of radiographic studies, we identified the deformities of the forearm bone, depending on the plane of their location. In all patients, regardless of the type of pathology according to the classification of Jo&d Jung, there was varus of the ulna and radius and recurrent deformity of the ulna. As for the severity of deformities in the forearm bones, according to the accepted values of the reference angles, the smallest deformities (regardless of the plane) were found in the third type. Also, with this type, we did not reveal shortening of the ulna with respect to the radius, and in patients with Type IVB this shortening was significantly bigger as compared with the patients of other groups (24.5 ± 11.9 mm).

The data obtained are presented in Table 2.

Table 1

Supination/pronation in patients with HMO, depending on the type of pathology according to the classification of Jo&Jung

Parameter	Type according to Jo&Jung			
	I	III	IVA	IVB
Supination (degrees)	62.3 ± 19.5	80.7 ± 8.5	37 ± 25.9	29.2 ± 25.9
Pronation (degrees)	73.9 ± 20.9	82.3 ± 19.6	55 ± 30	47.3 ± 34.3

Table 2

Forearm bone deformities in patients with HMO, depending on the type of pathology according to the classification of Jo&Jung

Parameter	Type according to Jo and Jung			
	Type I	Type III	Type IVA	Type IVB
Varus of the ulna (number of patients/%/deformity in degrees)	34/92/8.4 ± 4.5	23/85/4.1 ± 2.6	64/90/9.1 ± 5.87	12/92.3/15 ± 8.3
Recurvatum of the ulna (number of patients/%/deformity in degrees)	30/81/10.4 ± 5.79	15/55.5/7.5 ± 6.2	61/86/14.1 ± 7.0	10/77/12.6 ± 5.45
Varus deformity of the radius (number of patients/%/deformity in degrees)	19/51/8.45 ± 5.55	4/14.8/2 ± 1.1	45/63.4/ 10.1 ± 9.4	8/66.7/ 11.2 ± 11.6
PrNSA of the radius(degrees)	94 ± 5.2	91.6 ± 6.7	94.5 ± 6.69	98.9 ± 65.5
DEDA of the radius (degrees)	67.3 ± 4.78	65.4 ± 3.99	63.6 ± 6.75	65 ± 11.6
Shortening of the radius (mm)	14.8 ± 8.4	none	14.4 ± 7.9	24.5 ± 11.9
RAA (degrees)	21.8 ± 35.7	20.4 ± 9.8	22.3 ± 5.11	20 ± 28.6
RB (degrees)	10.2 ± 0.8	7.8 ± 2.1	8.7 ± 1.67	9.5 ± 28.6

Thus, the deformities most frequently encountered were varus deformity at the border of upper and middle thirds and recurvatum at the border and middle thirds in the ulna while in the radius the most common was varus deformity in the middle and lower thirds as well as dislocation or subluxation of the radius (30 %). We found that varus deformity of the radius in the middle third was absent in the cases with radial head dislocation (11 %). There was a combination of the forearm deformity variants mentioned above such as varus recurvatum deformity of the radius at the border of the upper and middle thirds (55 % of cases), varus recurvatum deformity of the ulna at the border of the upper and midle thirds combined with varus of the radius in the middle third (15 % of cases).

RAA and RB values were not significantly different in the patients with forearm deformities according to Jo & Jung's classification.

DISCUSSION

Long bone deformities have been sufficiently studied, as reported in domestic and international scientific journals. The authors mainly focus on the study and correction of long bone deformities of the lower extremities. The reference values of the angles of the ulna and radius and of epidiaphyseal angles of the radius were proposed after having conducted numerous retrospective analyses of radiographs [6, 20, 21].

Also, 213 cases of multi-planar deformities of long bones (of which only 7 cases were patients with a diagnosis of "Multiple hereditary exostoses") were described [22]. Deformity severity at the preoperative stage was assessed using the reference lines and angles.

A few articles deal with forearm deformities, including the deformities of the forearm in this genetic disease of multiple hereditary [23-25].

The researchers focus on the investigation of such parameters of the forearm deformities in MHO as ulna shortening, presence/absence of radial head dislocation, location of exostoses in the metaphyseal bone part of the forearm bones. So, N.D. Clement and D.E. Porter stated that the magnitude of ulna shortening relative to the radius and the radial head dislocation are the risk factors of rotation limitation of the forearm bones and note the need of early surgical interventions in children [26–28].

Most of the authors opine that the indication for deformity correction in MHO is ulna shortening by 1.5 cm or radius shortening by more than 60 %, increase in RAA more than 30°, radial head instability and restrictions of motion in the wrist and elbow joints [6, 16]. Ulna shortening averaged 14 mm in our patients that had types I и IVA of the deformities, and in type IVB it was up to 24.5 ± 11.9 mm [26–30].

A.R. Jo and S.T. Jung proved statistically in their work that the proportion of ulna shortening to the radius as well as forearm bone bowing (RB) were predictive factors of radial head dislocation [7]. We did not reveal statistical significance of the RB angle value even in the group of patients with radial head dislocation. The RB angle was smaller in the group of patients with osteocartilaginous exostoses located in the distal radius without dislocation and radius shortening.

CONCLUSION

The variety of forearm deformities in children due to HMO requires a differentiated approach to the choice of an individual, in each case, surgical treatment technique. It is important to clearly establish the indications for surgical intervention, depending on the deformity type and the specific clinical situation. To date, this issue remains unresolved and much discussed.

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Ethical Expertise *Patients and their representatives gave their consent to processing and publication of personal data.*

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