Literature review

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Forearm deformities in children with hereditary multiple exostosis (review of literature)

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Introduction Hereditary multiple exostoses, according to different authors, account for 16.0 to 43.0 % of all the cases of tumors, tumor-like and dysplastic diseases of the pediatric skeleton. Deformations of the forearm develop in 30–80 % of patients with multiple exostoses. **The purpose** of our research was a comprehensive presentation of the deformities of the forearm in children that occur in multiple exostoses, assessment of the efficiency of various surgical treatments. **Methods** We carried out the analysis of scientific medical articles reflecting the options of surgical treatment of this pathology (PubMed database) published between 1984 and 2018. The retrospective analysis of Russian-speaking and English-speaking sources of literature shows that elimination of forearm deformities in hereditary multiple exostoses is performed only in the surgical way. **Results** Analysis of results of surgical treatment established that 649 interventions published in literature resulted in 5.5 % of excellent, 33.5 % good; 23 % fair and 38 % poor outcomes. The most perspective and widely applied technique is gradual lengthening of the radius as an independent method of treatment, and in combination with other options of surgical interventions. **Discussion** Deformity recurrence and exostosis in most cases occur in patients with unfinished bone growth. The authors find optimal perform resection of exostosis in combination with other methods of treatment. At present, the dependence of the choice of surgical option on the severity of anatomical changes has not been established. There are no accurate indications to the choice of a specific technique of surgical intervention based on the deformity revealed. Despite improvement of treatment methods, the rate of poor results remains high and reaches 38 %.

Keywords: hereditary multiple exostosis, deformations of forearm, ulnar clubhand, children, options of surgical treatment

INTRODUCTION

As reported, osteochondromatosis occurs in 16.0 to 43.0 % of all cases of tumors, tumor-like and dysplastic diseases of the pediatric skeleton. The first descriptions of osteocartilaginous exostoses as a disease date back to 1740 [1, 2, 3].

Thirty years later, Boyer published the first description of a family case of multiple exostoses. This disease has a wide polymorphism of clinical manifestations, one of which is forearm deformities of varying severity. The incidence ranges from 30 to 80 %. A common location of osteocartilaginous exostoses on the bones of the forearm is the distal metaphysis of the ulna. In more than 80 % of cases, this leads to ulnar clubhand of varying severity [1].

This retrospective analysis of literature sources revealed that dysplastic and hereditary theories were most prevalent causes of osteochondromatosis. Researchers who adhere to dysplastic theory tend to consider osteochondromatosis as an anlage and developmental defect of the epiphyseal cartilage. This theory is more characteristic for solitary osteocartilaginous exostosis. Inherited theory has the largest number of supporters, it is mainly characteristic of multiple exostoses [3, 4].

The hereditary nature of multiple exostosis (ME) was established in the mid-nineteenth century. Targeted genetic studies have established the fact of hereditary transmission (by autosomal dominant type) more often through the male line. According to sources of foreign literature, genes 8, 11 and 19 were identified in which pathological changes are localized [5–9].

According to literature, 30–87 % of patients with ME develop forearm bone deformities. At the same time, deformities of the forearm due to solitary osteocartilaginous exostoses of the forearm bones are very rare, and the incidence of their occurrence was not found in the literature.

Clinical manifestations

An isolated lesion of the ulna is frequently manifested by a tumor-like formation in the area of its distal metaphysis. In some cases, first symptoms of the disease are dysfunctions of the joints: flexion contracture of the elbow joint, restriction of the rotational movements of the forearm, ulnar club

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hand. Deformities of the forearm bones, decentration, subluxation or dislocation of the radial head develop as the ulna shortening progresses [1].

In case both bones of the forearm are involved, shortening of the forearm, deformities of the forearm bones of varying severity, restriction / absence of rotational movements (in the presence of osteocartilaginous exostoses growing towards the opposite bone; subluxation, dislocation of the radial head), excessive ulnar deviation of the hand in the wrist joint are most often observed [9, 10].

Deformations of the forearm in children due to ME are, as a rule, progressive in nature during their growth period and worsen over time, causing dysfunction and disorders of elbow and wrist joint anatomy [11, 12].

Classification

In the Russian literature, the first classification that reflects shortening and deformities of the forearm bones and the condition of the wrist was published by V.A. Morgun in 1973. Later, it was expanded by A.P. Pozdeev and L.Yu. Khodzaeva. This classification is based on the degree of ulnar deviation, restriction of movements in the adjacent joints, presence or absence of forearm deformities, the magnitude of forearm shortening, presence / absence of restriction of rotational movements and decentration / subluxation / dislocation of the radial head. The authors presented five grades of ulnar club hand [12, 13].

Grade I: ulnar deviation of the hand within 10–15°; the wrist is actively and passively brought in the middle position, the range of motion in the adjacent joints is fully preserved.

Grade II: ulnar deviation of the hand is up to 30°; the wrist is actively and passively brought in the middle position. There is an arch-like deformity of the forearm bones; its shortening is up to 2.5 cm, restriction of rotational movements due to decentration and subluxation of the radial head.

Grade III: ulnar deviation of the hand more than 30°; the wrist cannot be brought to the middle position; deformation of the forearm bones and shortening is more than 3 cm, sharp restriction of rotational movements due to subluxation or complete dislocation of the radial head.

Grade IV: ulnar deviation of more than 30°, the hand cannot be brought to the middle position; there is a complete dislocation of the radial head, deformity of both forearm bones, rotational movements are within 10-15°.

Grade V: ulnar deviation of the hand is not more than 30°, lesions in the distal parts of both bones, dislocation of the radial head, deformity of the metadiaphyseal parts of both bones of the forearm, significant shortening of the segment, muscle hypotrophy, flexion contractures of the fingers.

In the English-language literature, the classification of the forearm lesions is also widespread, based on the localization of osseocartilaginous exostoses and the presence / absence of dislocation of the radial head [14] (Fig. 1):

Type 1: osseocartilaginous exostoses are located in the distal ulna;

Type 2A: dislocation of the radial head due to osseocartilaginous exostosis of the proximal radial metaphysis;

Type 2B: osseocartilaginous exostoses are located in the distal ulnar in conjunction with a dislocation of the radial head;

Type 3: osseocartilaginous exostoses are located in the distal radius.

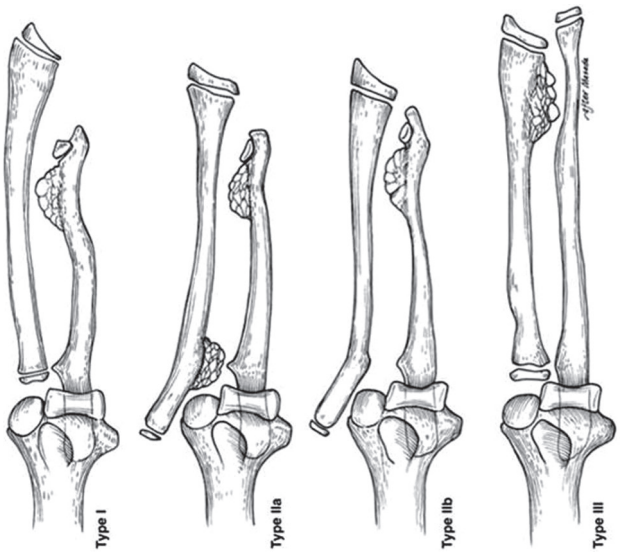

Fig. 1 Pathology variants according to K. Masada (1989)

Diagnosis

The main method for diagnosing forearm deformities is radiological study: X-rays of the forearm performed in two orthogonal projections (anteroposteior, lateral); computed tomography for better visualization of the nature of the deformity of the forearm bones. In addition, electroneuromyography is performed to assess the functional state of the neuromuscular apparatus, both before surgery and in the long term.

MATERIAL AND METHODS

The retrospective analysis of domestic and foreign literature sources (PubMed information database) found that the only effective method of forearm deformities treatment is surgical interventions.

From the reports published, the following surgical treatment options can be distinguished:

1) simple resection of osseocartilaginous exostosis;

2) distraction osteosynthesis (application of an external fixation apparatus);

3) internal osteosynthesis;

4) bone auto- or alloplasty;

5) temporary hemiepiphysiodesis.

Indications for surgical treatment are:

– osseocartilaginous exostoses that cause neurocirculatory disorders, accompanied by pain, functional limitations;

– club hand, accompanied by decentration, subluxation or dislocation of the radial head;

– disorders of the anatomical relationships in the adjacent joints;

– shortening of one of the bones of the forearm by more than 1.5 cm and shortening of both bones of the forearm by more than 4 cm as compared with an unaffected side [12].

We selected a number of medical articles on this pathology (from the PubMed information database) published between 1984 and 2018. Their authors present various combinations of surgical treatment methods. In the majority of cases, transosseous osteosynthesis was performed by applying a monolateral external fixation apparatus (Orthofix) to the ulna / radius alone or in combination with resection of osseocartilaginous exostosis / bone osteosynthesis / temporary hemiepiphyseodesis of the distal radius of the radius (in children before bone growth completion) in order to restore the distal epiphysiophysial angle and the relationship in the wrist joint. A retrospective analysis revealed that the most popular methods of surgical treatment are gradual elongation of the ulna with a monolateral external fixation device (Orthofix) [15–28, 46], resection of osseocartilaginous exostoses [24, 27–34], corrective osteotomy of the radius [17, 19, 22, 24, 25], temporary hemiepiphyseodesis of the distal radius growth zone [23, 32, 34, 35].

In multiplanar deformations of long tubular bones, it is necessary to use the method of transosseous osteosynthesis in combination with an apparatus based on passive computer navigation Ortho-SUV. The authors retrospectively selected 213 patients, whose average age was 12 years (of which 7 (3.2 %) were patients with multiple osteochodromas). According to the results obtained by the authors, this method was highly accurate in deformity correction [39].

The average at surgery, according to the authors, was the age of 9 years. However, there are two points of view regarding the age of surgical treatment. Some authors advocate early initiation of surgical treatment (in 4-5 year old children), since this contributes to further remodeling of the deformity of the forearm bones and a smaller volume of future surgical interventions [15, 19, 21, 26, 27, 28, 36, 39, 40, 41, 43].

Other authors do not agree with this point of view; they believe that it is most advisable to start surgical treatment before bone growth completion. They explain that the least number of surgical interventions and absence of further relapse of the deformity are thus expected. But this point of view has its minuses as long-existing and progressive deformities of the forearm lead to neuromuscular system and blood flow disorders, and gross violations of the relationship of the articular surfaces in the adjacent joints [20, 22, 24, 25, 30, 32, 34, 42, 45].

RESULTS

This retrospective analysis evaluated the efficiency of various surgical options according to the data of scientific medical articles (Pubmed information database) published from 1984 to 2018. The analysis of the results of various surgical treatment options found that out of 649 surgical interventions excellent results were obtained in 5.5 % of cases, good in 33.5 %; fair in 23 %, and poor in 38 % of cases. The group of "poor" results includes all relapses or unresolved deformities of the forearm, none increase in the amplitude of movements in the adjacent joints, and pain in the postoperative period. Of all the proposed surgical treatment methods, the most widely used was the gradual elongation of the ulna with AEF autonomously or in combination with resection of the osseocartilaginous exostosis, osteotomy of the radius (plate fixation). The most common complications were delayed consolidation / nonunion; fracture at the level of bone callus.

DISCUSSION

Relapses of deformities, as well as of exostosis, in most cases occurred in patients with uncompleted bone growth. Isolated resection of osseocartilaginous exostoses may be an auxiliary treatment with the aim of eliminating pain, cosmetic problems and, sometimes, to improve the range of movements. At present, there is insufficient evidence of spontaneous resolution of deformities or recovery of full range of motion when this method is used independently. The authors opine that it is optimal to perform resection of exostoses in combination with other treatment methods. Such methods as temporary hemiepiphyseodesis of the distal radius; fibula graft transfer on the vascular pedicle (on the ulna); one-bone forearm operation; resection of the radial head (in dislocation) in combination with autoplasty of the ligamentous apparatus; the osteotomy of the radius or its lengthening as an independent method has not been widely spread in order to draw objective conclusions. These surgical interventions can be applied in specific clinical situations, depending on the preferences of the surgeon.

Yuchan Li and co-authors state that interventions in children older than 12 years of age using a monolateral external fixator and performing an osteotomy in the metaphysical region allow for faster consolidation [22].

Kousuke Iba and co-authors, in turn, note that patients with a dislocation of the radial head need surgical treatment as soon as possible, since timely elimination of the dislocation with correction of the deformity and elongation of the ulna will not require further correction of the deformity of the radius. All patients (n=3) showed an increase in rotational movements (supination), had no pain, and a good cosmetic result was achieved [24].

Clement and Porter also advocate early surgical treatment in dislocation of the radial head, before the end of bone growth, since persistent dislocation leads to significant functional limitations, sometimes irreversible [25].

Arms *et al* conducted a survey of a group of 37 patients who underwent resection of the osseocartilaginous exostoses in the forearm bones. They found that patients did not experience functional limitations and were satisfied with their arm appearance, despite the deformity of the forearm. Surgical interventions improve the appearance and reduce pain when performed before or after the end of bone growth [36].

Peterson points out that deformity of the forearm bones is the most common cause of functional disorders due to ME. Thus, measures should be taken to prevent the progression of deformity and functional impairment as soon as it becomes known that the growth zone is affected [42].

Stanton *et al* evaluated the radiological parameters and dysfunction grade in a group of patients (n = 28) with multiple exostoses who did not undergo surgical treatment, and concluded that in most cases there was a good range of motion, despite the deformity. At the time of the examination, the average age of the patients was 21 years. On average, 5 % of the examined patients had functional disorders in the upper limb [44].

Akita S and Hill RA opine that surgical treatment in patients with radial head dislocation should be performed after the end of bone growth, since in most cases early surgical treatment leads to relapse of radial head dislocation [27, 31].

Wood *et al* reported a group of 10 patients who underwent surgery, such as lengthening, corrective osteotomy of the forearm bones, and indicated that despite a minimal functional improvement, the appearance of the limb improved noticeably [45].

Rozbruch *et al* reported the results of the use of a hexapod fixator in the correction of forearm bones. In all cases (n=5), deformity of the forearm bones was gradually eliminated; in one case, the radial head was reduced using a hexapod frame without neurocirculatory complications [47].

CONCLUSION

We conclude that gradual lengthening of the ulna, as an independent method of treatment or in combination with other surgical options, restores or improves the radiographic, clinical and functional parameters of the forearm. Excellent treatment results with a significant increase in the range of motion, absence of pain, and improvement of radiographic parameters were obtained in 5.5 % and good in 33.5 % of the cases reported. In all the cases, gradual elongation of the ulna was performed in combination with resection of the osseocartilaginous exostoses and reduction of the radial head. Treatment started at the age of 5 to 6 years. Despite a number of studies advocate surgical interventions upon the

end of bone growth or its rejection, the results of which are statistically insignificant, the age of 5 to 6 years is considered the most suitable for surgical interventions. To date, no classification has been developed for variants of multiplanar forearm deformities and the corresponding treatment tactics based on the nature of the deformity and the age of a patient. A clear dependence of the choice of surgical options on the severity of anatomical changes has not been established. There are no clear indications for selection of a specific surgical procedure, taking into account the deformity type. Despite the improvement of treatment methods for this pathology, the rate of poor results remains high and reaches 38 %. To date, these issues remain the subjects of numerous discussions.

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