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Peculiar features of orthopaedic and somatic condition in patients with severe types of cerebral palsy

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I**ntroduction** Hip dislocation, accompanied by pain and segment malposition that affects the sitting posture and excludes passive verticalization, has been recognized as the main cause that worsens the quality of life in patients with severe types of cerebral palsy (GMFCS levels IV and V) with unreconstructable hip dislocation. **The purpose** of this study was to assess the orthopedic status of patients with spastic types of cerebral palsy that correspond to GMFCS functional levels IV and V with dislocated unreconstructable hip, as well as to examine their somatic status at admission for operative orthopedic treatment. **Material and methods** A continuous series of patients (41 patients, mean age 14.1 ± 2.3 years) with severe types of cerebral palsy that were admitted for palliative surgical treatment of femur dislocation and associated contractures and deformities in lower limb segments was studied. Orthopedic and somatic status was examined; radiological studies of the hip, knee joints, and feet were conducted. **Results** Changes in the femoral and acetabular components of the hip joints, detected with X-rays and CT, showed a nonreconstructable hip and confirmed the indications for palliative interventions. Clinically significant contractures and deformities in the knee joints and feet that required surgical treatment were present in 61 and 70.7 % of cases, respectively to levels. In this series of patients, concomitant diseases were found in all cases. Deficits of body mass index were present in 41.7 % of cases with GMFCS level IV and in 76.5 % of cases with GMFCS level V. **Conclusion** Severe pain and lower limb malposition is the leading symptomatic complex in patients with severe types of cerebral palsy of GMFCS levels IV and V that need correction. X-ray and CT findings in our series testified unreconstractable hip joint. In most cases, there were clinically significant deformations and contractures of the knee, ankle and foot that required a simultaneous correction. Thus, surgical interventions in such situations should be performed at several anatomic levels according to the principles of one-stage multilevel intervention. All patients in the study group had concomitant diseases and bone mass index deficits were observed. The situation with regard to concomitant diseases and body weight deficiency is more unfavorable in patients with GMFCS level V.

Keywords: cerebral palsy, unnreconstructable hip dislocation, severe motor disorders, one-stage multilevel orthopedic surgery

INTRODUCTION

The most frequent and serious orthopedic complications in patients with spastic types of cerebral palsy (CP) is hip dislocation which is seen in 30 to 63 % of cases [1–4]. From the age of 10 years and older, reconstructive treatment for hip dislocation in CP patients with functional levels IV and V according to the GMFCS scale [5] is considered unjustified in most cases due to severe deformities of the femoral head and acetabulum, accompanied by loss of articular cartilage and underlying bone tissue [1, 6, 7]. Painful dislocation of the hip, segment mal-position that affects correct sitting and does not allow passive verticalization, is the main cause that worsens the quality of life and severely limits functional ability of such patients [2, 8, 9]. In addition, CP patients frequently have chronic pathology of the digestive, cardiovascular, respiratory and urinary systems as well as a serious decrease in the body mass index (BMI) [10–18].

The goals of palliative operative orthopedic treatment in such patients are pain elimination or relief, correction of lower limb mal-position, creation of the possibility of unhindered mobilization of the lower limbs and passive verticalization, and improvement of the conditions for patient's care [19–22].

Associated foot deformities, contractures of the knee joints are also the factors that impede patient's verticalization, comfortable sitting posture and wearing of shoes, and worsen the quality of life [23–25]. We believe it is important to correct simultaneously the deformities of the entire lower limb biomechanical axis in order to achieve the maximum possible result [22]. But it is also important to consider poor somatic conditions of patients with severe CP types while planning operative correction.

The purpose of this study was to assess the orthopedic status of patients with CP spastic forms, corre-

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sponding to functional levels IV and V according to the GMFCS scale, complicated by unreconstructable hip dislocation, and also their somatic status at admission for operative orthopedic treatment.

MATERIAL AND METHODS

We conducted a study of orthopedic disorders and somatic status of a continuous series of patients (41 patients, spastic CP types, IV and V levels according to the GMFCS scale, M/F = 23/18) who underwent orthopedic surgical treatment for hip dislocation at the RISC for RTO in the period between 2012 and 2016. The average age at the time of admission was 14.1 ± 2.3 years (range: 10 to 18 years).

Inclusion criteria were age of 10 years and older, spastic CP types complicated by a unreconstructable dislocation in the hip for which palliative interventions were performed. Patients with other neuromuscular diseases and CP patients who previously underwent operative reconstructive treatment for hip dislocation were excluded.

The overall motor activity of children was assessed by the GMFCS system [5]. Orthopedic assessment included the study of the range of the hip, knee and ankle joints motion, lower limb deformities and deformations of the spine. Pain intensity was assessed by the face assessment scale [26] at rest, during normal daily movements and during the sessions of kinesitherapy. The possibility of passive verticalization with loading the lower limbs, the possibility of giving a symmetrical comfortable posture while lying down or sitting, the convenience of performing hygiene procedures was examined. Parents or their care persons clarified about bedsores and other trophic changes in soft tissues due to a monotonous posture.

Radiographically, such indicators as the Reimers index, the projectional neck-to-shaft angle (NSA), the acetabular index (AI), the Sharp's angle, the sphericity of the femoral head, the concentricity of the head and the acetabulum, the magnitude of the femoral head displacement above the acetabulum, the index of the acetabulum depth were calculated [27, 28]. Computed tomography of the hip joints performed with the Toshiba Aquilion-64 system (Japan) was used to assess acetabular anteversion, the axial acetabular index, the CT-acetabular index, predominant location of the expressed acetabular dysplasia, femoral neck anteversion, the true NSA, and to measure the size of the bone defect of the head and its location [29–32]. Among the numerous X-ray measurements for the knee and foot study, the Caton-Deschamps index (index of the patella height) was chosen [33, 34], the tibiotalar angle, the angles between the axes of the first metatarsal bone and the proximal phalange examined on foot AP and lateral radiographs, as well as the angle between the axis of the first metatarsal bone and the axis of the talus examined in the lateral views [35-37].

All patients were consulted by pediatricians or therapists before surgery. When assessing somatic status and concomitant pathology, clinical history, physical examination, standard anthropometric parameters (height, weight, BMI), ECG results, laboratory test results (total blood and urine counts, blood biochemistry, coagulogram); data obtained with additional methods and the results of specialist consultations were considered. As for BMI, we opine that the normal limits of this parameter for patients with severe CP types should differ from those in a healthy child population [38, 39]. So, pathological BMI values are below the 10th percentile of the values for a healthy population [10, 40].

The quantitative data obtained were subjected to statistical processing using Microsoft Excel 2016 software. The statistical study included descriptive statistics: mean values (M) and standard deviation (SD).

RESULTS

In this series, GMFCS level IV of motor disorders was diagnosed in 24 cases and 17 patients had level V. **Table 1** presents the mean values of age, weight, height, BMI according to the functional levels.

It should be noted that ten of 24 patients (41.7 %) with functional level IV had the BMI below the 10th percentile. In children with more severe and extremely severe motor and neurological disorders corresponding to GMFCS level V, the BMI below

the 10th percentile was found in 13 cases (76.5 %).

For the whole group, the pathology of the cardiovascular system was congenital heart disease in three cases; sinus arrhythmia, sinus tachycardia and conduction disruption, one case of each. Diseases of the eyes were found in 18 patients: partial atrophy of the optic nerves (6 cases), strabismus (9 cases), myopia (2 cases), and anisocoria (1 case). Diseases of the gastrointestinal tract were biliary dyskinesia (2 cases), constipation (23 patients), incontinence of the feces (1 case), and functional indigestion was found in 1 patient. Anemia of mild severity (hemoglobin 90-110 g/l) was present in 8 cases. Hydrocephaly was found in two children, microcephaly in 4 cases, epilepsy was present in 10 patients and bulbar disorders and aphasia were detected in one case. Problems with the control of urination were reported in five patients. The endocrine system disorders were congenital hypothyroidism without goiter in one case and decrease in bone mineral density, confirmed by the data of absorption densitometry, in two cases. In general, each patient had some type of concomitant pathology. Most of concomitant diseases (54 instances, 65.9 %) were detected in GMFCS level V patients (**Table 2**).

Changes in the values of blood biochemical tests beyond the limits of the reference values of the age-related physiological norm were detected in 12 patients: hypocreatinemia (4 cases), hypobilirubinemia (3 cases), hyperalbuminemia (2 cases), hyperglycemia (1 case), hypoglycemia (1 patient), and increase in the level of aspartate aminotransferase (1 case).

Orthopedic complications of cerebral palsy in the lower extremities were neglected hip dislocation (bilateral in 23 cases and one-sided in 18 cases), accompanied by typical degenerative and destructive changes in the femoral head and acetabulum revealed by image studies.

These changes were accompanied by a pain (level 3 and higher on the scale of facial pain assessment) at rest in the maximum possible comfortable position in 38 patients; by daily routine movements, during hygienic procedures and during kinesitherapy sessions in all patients (41 cases).

For all cases, representatives of patients pointed out serious difficulties in performing hygienic care in the perineal region due to pronounced femur flexion and adduction. All patients showed asymmetrical sitting and lying postures. The postural management was extremely difficult. There was no possibility of passive verticalization with support on the lower limbs (**Fig. 1**). Trophic changes in the area of greater trochanters and femoral condyles due to the monotonous posture were reported in the anamnesis in 19 cases.

Data from imaging studies confirm the severity of pathological changes in the hip joints (**Tables 3, 4**; **Fig. 2** and **Fig. 3**).

Table 1

Mean values of age, weight, height, BMI

	Age (years)	Height (cm)	Weight (kg)	BMI
GMFCS IV (n = 24)	13.6 ± 2.3	141.2 ± 8.6	37.6 ± 11.2	18.6 ± 4.0
GMFCS V (n = 17)	14.5 ± 2.4	138.2 ± 10.8	25.3 ± 5.9	13.1 ± 2.5

Table 2

Concomitant pathology distribution according to GMFCS levels

	GMFCS IV	GMFCS V
Cardiovascular system	1	5
Digestive system	12	14
Urinary tract system	1	3
Hemapoietic system	4	4
Vision system	5	13
Central nervous system	5	12
Endocrine system	–	3
Total	28	54

Fig 1 Photos of patient P., 13 years old, before the treatment: **a** – spontaneous monotonous posture while patient is lying on the spine; **b** – position of limbs and trunk by an attempt to passively verticalize for sitting and standing; **c** – maximum possible femur abduction and extension of the left knee joint

Table 3

Radiographic coxometry indicators

	Reimers index (%)	Projectional NSA, degrees	Sharp's angle, degrees	Distance between the supposed centres of the head and acetabulum, mm	Magnitude of the proximal shift of the femur, mm	Acetabular depth index (%)
GMFCS IV	91.6 ± 17.2	158.9 ± 9.2	51.9 ± 5.3	44.4 ± 8.7	20.4 ± 13.5	17.9 ± 5.6
GMFCS V	93.0 ± 20.8	160.0 ± 16.0	49.1 ± 5.2	42.1 ± 9.5	22.1 ± 14.3	23.0 ± 8.9

Table 4

CT indicators of the hip joint

	Acetabular anteversion, degrees	Axial AI, degrees	Frontal AI, degrees	Femoral neck anteversion, degrees	True NSA, degrees	Longitudinal and transverse sizes of the defect of the head, mm	Head defect depth, mm
GMFCS IV	25.4 ± 10.7	119.2 ± 13.9	36.6 ± 5.7	41.8 ± 19.3	141.7 ± 8.6	21.0 ± 5.0 / 22.2 ± 9.8	5.2 ± 2.5
GMFCS V	22.7 ± 6.8	114.9 ± 15.9	30.1 ± 7.4	39.4 ± 17.7	135.8 ± 9.4	19.4 ± 8.0 / 19.5 ± 5.5	4.6 ± 2.2

Fig 2 AP radiographs of the pelvis in patients with severe cerebral palsy (GMFCS levels IV and V) illustrating the typical deformities of the femoral head and acetabula: **a** – right-side dislocation: Sharp's angle is 52° (right) and 40° (left), index of the acetabular depth is 9.5% (right) and 44.4% (left); **b** – bilateral dislocation: Sharp's angle is 55° (right) and 51° (left), acetabular depth index is 24.7% (right) and 26 % (left)

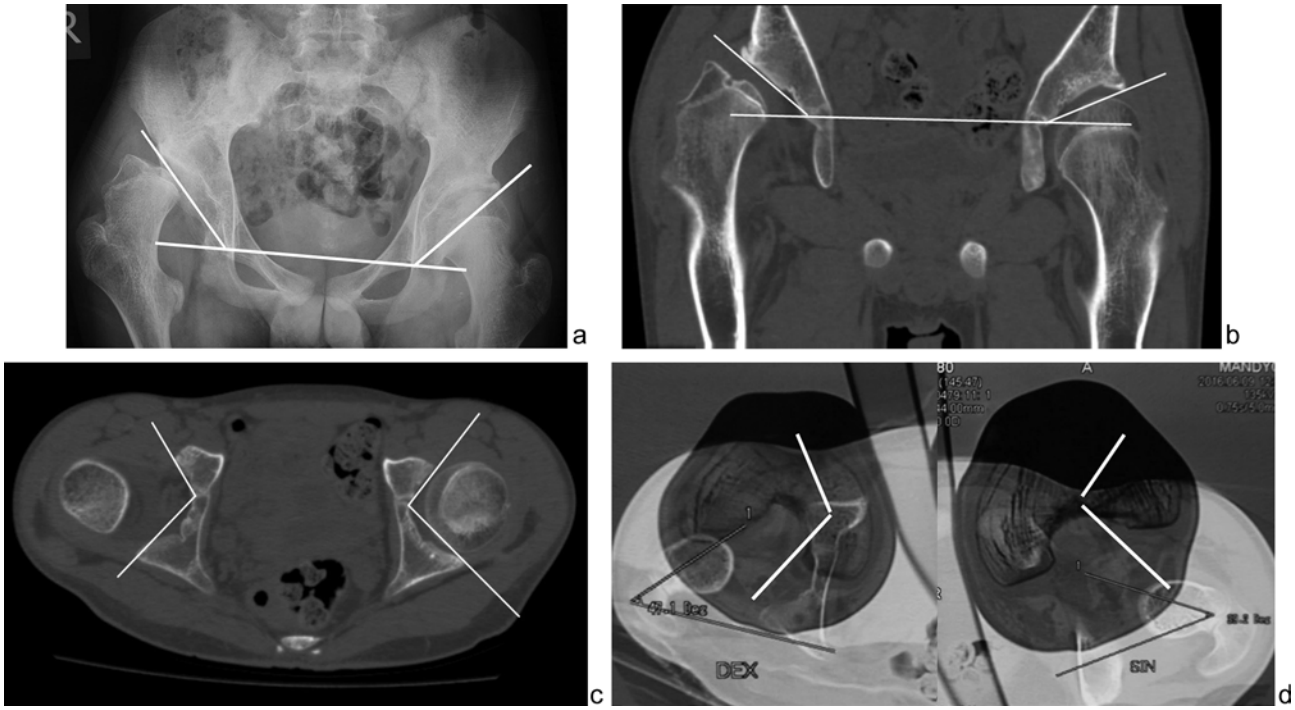

Fig 3 AP radiographs of the pelvis and CT images of a 15-year-old patient with GMFCS level IV: **a** – radiograph of the pelvis, right-sided dislocation: Sharp's angle is 52 ° (right) and 46 ° (left); **b** – frontal section: AI is 41 ° on the right and 25°on the left; **c –** axial section: axial acetabular index of 109° on the right and 96° on the left; **d** – angles of anteversion of the femoral necks (right 47°, left 39°)

Radiographic findings, obviously, reflect not only the fact of dislocation of the femur, but also severe changes in both the femoral and acetabular components of the hip joint.

Computer tomography data also reveal serious anatomical disorders in the proximal femur and acetabulum. Both imaging methods showed a pronounced disorder of the femoral sphericity, nearly a complete loss of its round shape, which in combination with the defect of the femoral head reflects the unreconstructable hip joint.

The expressed knee joint flexion (deficiency of passive extension 120°–30°) was present in 25 patients (31 joints). Comfortable sitting posture was impossible in patients with flexion of 90° (7 cases) or more. In 14 cases, the flexion contracture of the knee joint was less pronounced (deficiency of passive extension 20°–0° with an active extension deficit of 30°–10°). In the remaining two patients, active and passive extension in the knee joint was full. The pathologically high position of the patella was observed in all the cases of flexion contracture of the

knee joint (Caton-Deschamps index: 1.41 ± 0.15).

Scoliotic deformity of various grades was present in all patients. There were indications for an operative correction of spinal deformity in five cases (thoracolumbar and lumbar scoliosis with Cobb's angle of more than 50˚), but a preliminary stage of surgical treatment was required to eliminate the pathological hip flexion-adduction position.

Severe foot deformities (flat valgus in 26 feet; calcaneal valgus in 28 feet) (**Fig. 4**) that impede the use of ordinary or orthopedic shoes, or valgus deformities of the first toes combined with flexion contracture in the first metatarsophalangeal joint (35 feet) were detected. Totally, 54 segments were affected in 29 patients. In most cases (42 feet), there were signs of arthrosis of the Chopart's joint (deformity of the head of the talus) and the first metatarsophalangeal joint (deformity and flattening of the head of the first metatarsal bone). **Table 5** presents the quantitative data of foot parameters, the deformities in which were of clinical significance.

Severe flat valgus was characteristic of patients with GMFCS level IV (20 feet); calcaneal valgus was dominant in level V patients (22 feet) or in patients with level IV motor disorders in whom the tendon of the triceps muscle had been previously excessively elongated (6 feet).

Finally, it is important to note that hip dislocation alone, without any associated orthopedic pathology, was not found in any patient (**Fig. 5**), which is due to the systemic nature of CP disorders.

Fig 4 Radiographs of knee joints and feet with orthopedic pathology: **a** – high patella, Caton-Deschamps index is 1.6 on the right and 1.7 on the left; **b** – lateral deviation of the forefoot (left), angle of the talocalcaneal divergence of 51°, angle between the axis of the talus and the axis of the first metatarsal bone of 62° on the left; valgus deviation of the first toe (right), angle between the axis of the first metatarsal bone and the axis of the proximal phalanx of 42°; **c** – variants of foot deformities detected on the lateral view (from left to right): angle between the axis of the first metatarsal bone and axis of the proximal phalanx equal to 40˚, angle between the axis of the talus and the first metatarsal bone of 34˚, tibiotalar angle of 26°

Table 5

Radiografic foot parameters

	Tibiotalar angle, degrees	Angle between the talus and 1st metatarsal, degrees	Angle betwee the 1st metatarsal and main phalange, degrees (AP view)	Angle betwee the 1st metatarsal and main phalange, degrees (lateral view)
Flat valgus	136.2 ± 14.6	79.2 ± 21.8	44.3 ± 22.7	45.7 ± 21.3
Calcaneal valgus	77.4 ± 18.2	28.3 ± 19.5	39.2 ± 18.4	74.5 ± 21.1

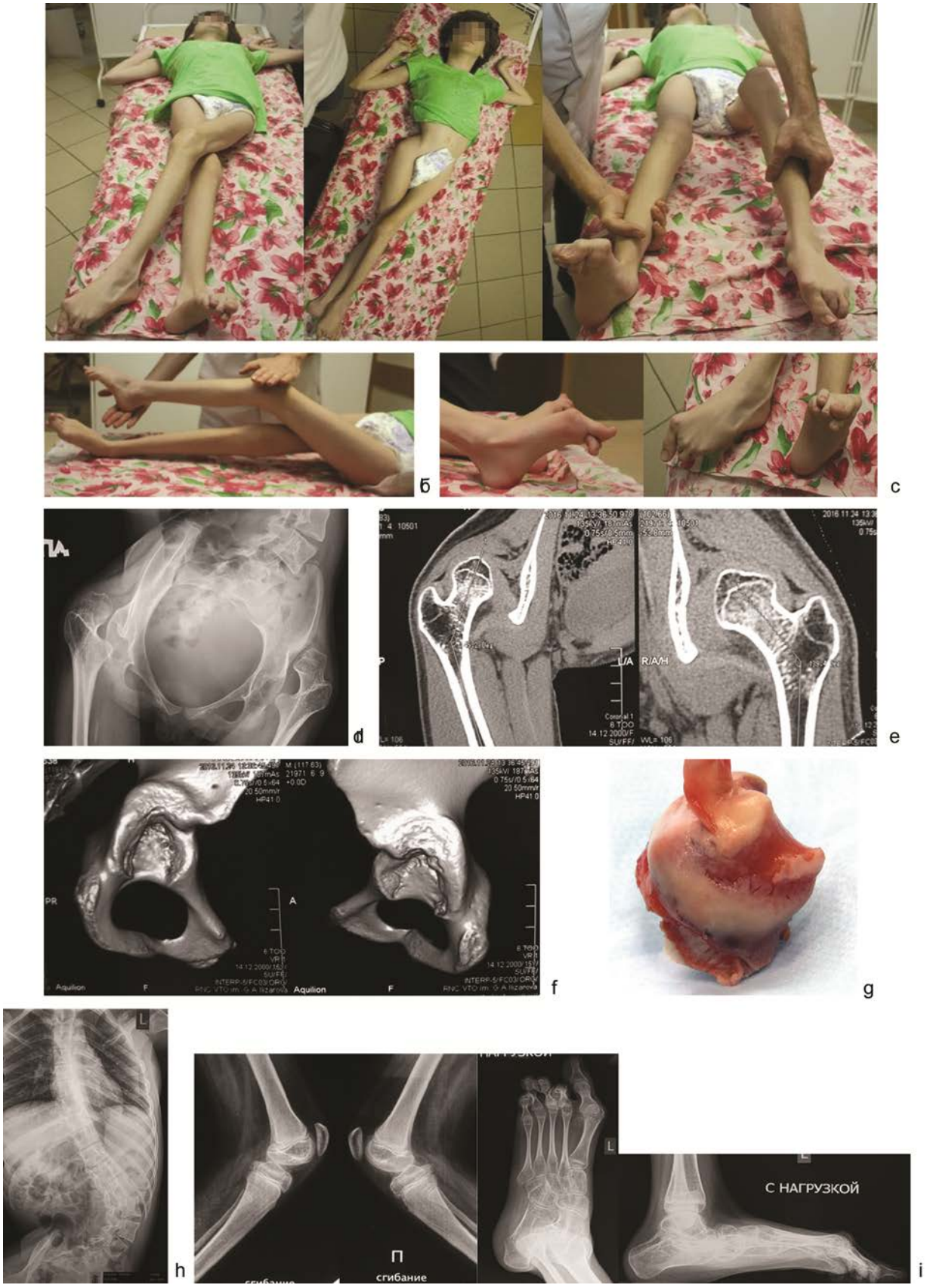

Fig 5 Deformities and contractures in patient P., 16 years old, GMFCS level V: **a** – expressed hip adduction contractures; **b** flexion contracture of the knee joint; **c** foot deformities, flexion contracture in the metatarsophalangeal joints; **d** pelvis adiograph, bilateral dislocation of the hips; **e** CT in the frontal plane illustrating the femoral head deformity and complete loss of sphericity; **f** 3D reconstruction demonstrates pronounced deformity of the acetabulum; **g** photo of the resected head of the left femur showing the loss of articular cartilage and extensive defect of bone tissue; **h** lumbar left-sided scoliosis, Cobb's angle of 87°; **h** radiographs of the knee joints (high patella) and feet (tibiotalar angle of 112 °, angle between the axis of the talus and axis of the first metatarsal bone of 31°, angle between the axis of the first metatarsal and the axis of the proximal phalange of 23 °

DISCUSSION

Hip dislocation in patients with spastic cerebral palsy, corresponding to GMFCS levels IV and V, is a frequent and serious complication that occurs in 30 to 63 % of cases [1-4]. Predisposing factors for lateralization and dislocation of the head are spasticity and retraction of the adductor group of the femoral muscles and iliopsoas muscle, excessive femoral neck anteversion, its valgus deformity, and lack of functional load on the limb [41-44]. These anatomical abnormalities were detected in all patients of the group.

The main problems that arise in CP patients with hip dislocation who are over 10 years of age are severe pain, uncomfortable sitting posture, impossibility of passive verticalization, difficulties in performing hygienic care of the perineal region, predisposition to pressure sores due to the monotonous posture, and difficulties in daily manipulations and actions (for example, changing clothes in a patient) [45–48].

Severe pain due to arthrosis in the hip joint is the leading problem among a number of numerous orthopedic problems in such patients. Its rate reaches 70 to 100 % [45–48]. Severe pain requiring correction with medications was found in 38 out of 41 patients (92.7 % of cases) at rest and in a comfortable position, while 100 % of cases experience it during movements in everyday life and during exercise therapy.

The femur flexion contracture is typical for this pathology and is encountered in all cases on the side of the dislocation. Independently, or along with pain, it is an obstacle to achieving a comfortable sitting posture and hygienic care [45, 47]. In our series, severe contractures and mal-position of the femur on the hip dislocation side were observed in all patients. Sufficient full-range flexion and extension of the femur was present only in unilateral dislocations in the intact hip joint.

Reconstructive interventions in patients of similar age and degree of neurological disorders as our group are considered unjustified, due to severe deformities of the femoral head and acetabulum accompanied by a subtotal loss of the articular cartilage and underlying bone tissue [2, 7, 9, 43]. Root [47] points out that the technical possibility of hip reconstruction still remains in older patients, but this procedure is not justified in terms of eliminating pain and restoring the possibilities of passive hip mobilization [46]. Van Riet and Moens explain the futility of reconstructive interventions in such patients by the lack of the potential for remodeling the deformed articular ends in the process of residual physiological growth [7].

Flexion knee joint contractures are also observed in patients with severe CP types, causing not only the complexity or even the inability to use verticalizers (by moderate irreducible joint flexion), but also the uncomfortable or total inability to acquire a sitting posture (by 90 or more degrees) [33, 36]. In our group, clinically significant contractures were present in 25 patients (61 %).

Foot deformities, which are clinically important even in patients unable to move independently, are the object of surgery if they make it difficult to use shoes or bring difficulties in foot skin care and contribute to trophic disorders [23, 35, 37]. Among our patients, surgical correction of foot deformities was required in 29 patients (70.7 %), but disorders were not always bilateral.

The need for passive verticalization of such patients with full weight-bearing on the lower extremities is extremely important in terms of improving the overall quality of life and preventing complications in the internal organs, as well as for preventing fractures due to low bone mineral density [11–18, 49, 50]. But to achieve the conditions for the location of patients, for example, in the posterior support verticalizer, simultaneous correction of deformities in the segments of the whole biomechanical axis of the lower limb is required. In our group of patients, it was necessary to intervene on several segments of the lower limbs at once to ensure comfortable verticalization and to improve the quality of life. Isolated intervention on the hip joint by associated pathologies of the knee joints and/or foot would not achieve comfortable conditions for patient care, kinesiotherapy and postural management.

However, the patients with severe CP types have concomitant somatic pathologies [11–18, 51]. In addition, such patients have deficient body weight with their BMI lower than the 10th percentile. It occurs in 36–51 % of the population of patients with the disorders corresponding to GMFCS levels IV and V [16, 38–40]. In their study, Ryabykh T.V. et al. [51] found that the BMI lower

than the 10th percentile as compared with the normal population was present in 49 % of cases with severe CP types and was combined with reduced hematopoiesis indices. In our group of patients, one or another (more commonly, a combination) concomitant pathology was present in each patient, but 65.9 % of associated conditions were detected in patients with GMFCS level V of motor disorders. Deficient BMI was present in 41.7 % of cases of GMFCS level IV patients and in 76.5 % of cases corresponding to the GMFCS level V, but only in 8 cases it was combined with mild anemia.

CONCLUSIONS

Our study confirmed that severe pain and lower limb malposition is the leading symptomatic complex in patients with severe types of cerebral palsy of GMFCS levels IV and V that need correction. Radiographic and CT findings in our series testified the impossibility of hip joint reconstruction. In most cases, there were clinically significant deformities and contractures of the knee, ankle and foot that required simultaneous correction for sitting posture comfort, adequate passive weight-bearing verticalization, convenience for kinesotherapy procedures, postural management, patient's hygienic care and improvement of quality of life. Thus, surgical interventions in such situations should be performed at several anatomic levels according to the principles of one-stage multilevel intervention. Concomitant diseases and frequent bone mass index deficits that feature all patients in this study group should be considered. The situation with regard to concomitant diseases and body weight deficiency is more unfavorable in patients with GMFCS level V.

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