

## ONCOLOGY

# Surgical hip dislocation according to Ganz for excision of osteochondromas in patients with multiple hereditary exostoses

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#### **Aims**

We report a prospective cohort study of the midterm results of surgical dislocation of the hip (according to Ganz) to perform resection of osteochondromas involving the femoral neck in patients with multiple hereditary exostoses (MHE).

#### Methods

Hip range of movement (ROM) was assessed pre- and post-operatively. Patients' judgment of post-operative reduction of pain, symptoms, the Rand 36-item Health Survey (RAND-36) and complications were analysed.

#### Results

Symptomatic osteochondromas of the femoral neck were removed in 20 hips (17 patients) between 2007 and 2012. There were nine men and eight women with a mean age at the time of surgery of 29 years (11 to 47). Mean follow-up was 46 months (26 to 73). At latest follow-up, mean ROM was significantly increased in all directions. Post-operatively the pain associated with the lesion was either significantly decreased or non-existent. There was a significant improvement in seven RAND-36 sub-domains. Encountered complications in four patients were pseudoarthrosis of the trochanteric osteotomy, traumatic separation of the trochanteric osteotomy, a pertrochanteric femoral fracture and avasvular necrosis. Histological analysis revealed osteochondromas in all hips.

# Discussion

This study confirms the Ganz trochanteric flip osteotomy provided a reliable approach to osteochondromas of the femoral neck that are otherwise difficult to access for surgical resection. The procedure offered significant improvement in the quality of life, although one should be aware of the serious complications can arise despite the relatively safe procedure.

Take home message: When daily function and activities are affected, resection of osteochondromas of the proximal femur according to Ganz is indicated to significantly improve quality of life.

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Multiple hereditary exostoses (MHE) or multiple osteochondroma (MO) is a monogenetic autosomal dominant disorder characterised by multiple osteochondromas developing at the metaphysis of long bones and the axial skeleton in childhood. <sup>1,2</sup> These osteochondromas of MHE may be associated with clinical problems including pain because of compression on the surrounding soft tissue, overlying bursal inflammation, impairment of articular function, cosmetic deformity and impingement of nerves, blood vessels and tendons. <sup>3-10</sup> Furthermore, limb deformities may arise, which are generally irreversible after skeletal maturity. <sup>2</sup> Surgical excision of symptomatic osteo-

chondromas may be necessary to improve joint function and reduce complaints of pain.

The most common locations for osteochondromas are the distal femur, proximal humerus and proximal tibia respectively. According to the literature, the proximal femur was involved in between 30% and 90% of patients with MHE. <sup>11,12</sup> However, we found the prevalence to be 80% in a cohort of 185 patients treated in our department. <sup>13</sup> Symptoms from proximal femoral osteochondromas are not as troublesome in patients with MHE as those found in other locations. Nevertheless they can lead to deformity of the femoral neck, resulting in pain

owing to mechanical restriction, functional impairment of the hip joint, bursal inflammation or femoroacetabular impingement as the widened and enlarged femoral neck comes into contact with the acetabular rim. <sup>10,14,15</sup>

Resection of intra- and periarticular osteochondromas of the hip might reduce pain and increase the range of movement (ROM) of the hip. 16-18 Theoretically, early surgical removal of osteochondromas, in particular those found involving the medial side of the femoral neck, could also reduce the risk of subluxation of the hip. 11,13,16 Since the anterior, inferior (medial) and posterior aspects of the femoral neck could be involved in patients with MHE, resection of the osteochondromas poses difficulties in choosing an adequate surgical approach. 15,19 The surgical approach as reported by Ganz et al<sup>20</sup> provide an unobstructed view of the femoral head, neck and intra-articular area and can therefore be used for excision of osteochondromas in this location.

The objectives of this study were to demonstrate the clinical outcomes and risk of complications of excision of oste-ochondromas of the femoral neck in patients with MHE using the Ganz approach to surgical dislocation of the hip.

#### **Patients and Methods**

Design and patients. This prospective cohort study was performed between 2007 and 2012 in the Onze Lieve Vrouwe Gasthuis, which is a reference centre for MHE in the Netherlands. A total of 17 consecutive patients with MHE and symptomatic osteochondromas of the femoral neck involving 20 hips were treated surgically. There were eight women and nine men with a mean age of 29 years (11 to 47) at the time of surgery. In the three patients with bilateral lesions, each hip was treated on separate occasions. In all 12 right hips were affected and eight left hips. Surgical resection was indicated for pain in two hips (10%), functional impairment in two (10%) or both in 16 (80%). Radiographs were made to localise and visualise the lesions and pre-operative MRI scans were carried out to assess for features of malignant transformation.

Surgical technique. All hip joints were approached as previously described by Ganz et al.<sup>20</sup> Briefly, the patients were operated in a lateral decubitus position. A Kocher-Langenbeck incision was made and the fascia lata was split longitudinally.<sup>20</sup> A greater trochanteric flip osteotomy was performed which ended in front of the posterior insertion of the gluteus medius to protect the branch of the medial femoral circumflex artery (MFCA). The greater trochanter was flipped anteriorly along with the vastus lateralis and the gluteus medius. The interval between the gluteus minimus and the tendon of the piriformis was developed and the capsule was exposed by retracting the gluteus minimus superiorly. After a Z-shaped capsulotomy, the hip joint could be anteriorly subluxed or dislocated to resect the osteochondromas. Cam lesions were removed to rectify insufficient offset of the femoral head-neck and labral lesions could be partially excised if indicated. The articular capsule was re-approximated and the trochanteric flip osteotomy was secured back to its bony-bed with two or three cortical screws. Following surgery, weight-bearing was not allowed for six to eight weeks. Patients received 100 mg of indomethacin immediately after surgery and 50 mg indomethacin three times daily until three weeks postoperatively to prevent the formation of heterotopic bone post-operatively.

Outcomes. Hip joint function was tested pre- and postoperatively in five principal directions by the surgeon at the outpatient clinic: flexion, external rotation, internal rotation, abduction and adduction, which were recorded to the closest 5°. Conventional anteroposterior and lateral radiographs were performed one day after surgery and during follow-up at the outpatient clinic. Upon a mean follow-up evaluation of 35 months, the patients completed the Rand 36-item Short-Form Health Survey (RAND-36)<sup>21</sup> evaluating eight health dimensions and two summary scores (physical component summary and mental component summary).<sup>21</sup> Responses could be transformed into a 0 to 100 range for each health dimension to evaluate pre- and post-operative results. Pre-operative scores were also compared with RAND-36 records of the general MHE population in the Netherlands.9

The post-operative mean reduction of pain was analysed with use of the numeric rating scores (NRS) for pain, in which 0 represents no pain and 10 represents worst imaginable pain.<sup>22</sup> Furthermore, patients were asked to grade their satisfaction with the outcome in a four-point scale (fully satisfied, partially satisfied, partially dissatisfied and completely dissatisfied).

Complications were evaluated until the most recent postoperative visit. MRI-scans for the evaluation of avascular necrosis (AVN) of the femoral head were performed in all patients with complaints first reported at the six-month follow-up.

**Statistical analysis.** A statistical analysis of pre- and postoperative ROM, NRS values and RAND-36 scores was performed with use of SPSS version 17.0 Software (SPSS Inc., Chicago, Illinois)). The Wilcoxon signed-rank test was used to assess the difference between pre- and postoperative records. Pre-operative RAND-36 scores were compared with reference RAND-36 scores of the general MHE population with use of the one sample *t*-test. Statistical significance was set at a p-value of < 0.05.

### **Results**

Conventional radiographs and MRI scans of the hip joints showed a typical MHE pattern of osteochondromas with pedunculated and/or broad-based lesions in all patients (Fig. 1). On pre-operative MRI scans, there was no suspicion for malignant degeneration of any of the lesions.

The mean length of hospital stay after surgery was 4.5 days (2 to 13). The mean post-operative follow-up was 46 months (26 to 73). Immediate post-operative radiographs (Fig. 2) did not reveal any complications and histopathological





Fig. 1a

Fig. 1b

a) Posteroanterior and b) lateral pre-operative plain radiographs demonstrating the typical oste-ochondrama of the femoral neck associated with Multiple Hereditary Exostosis.



Fig. 2

Post-operative posteroanterior plain radiograph (same patient as Fig. 1) after resection of an osteochondroma using surgical dislocation of the hip according to Ganz et al.<sup>2</sup>

Table I. The pre- and post-operative ranges of hip movement (°) in 17 patients (20 hips) with multiple hereditary exostoses, which was assessed with use of the Wilcoxon signed-rank test

	Pre-operative	Post-operative	p-value
	Mean (SD)/median (IQR range)	Mean SD/median (IQR range)	
Flexion	89.8 (18.2)/ 90 (80 to 100)	109.0 (13.5)/110 (100 to 120)	0.001
External rotation	16.3 (16.0)/10 (7,5 to 22,5)	36.7 (13.1)/32.5 (30 to 45)	0.001
Internal rotation	15.5 (14.4)/17.5 (0 to 30)	34.4 (12.9)/35 (30 to 40)	0.001
Abduction	37.0 (14.0)/40 (30 to 45)	47.8 (10.2)/50 (40 to 50)	0.024
Adduction	11.5 (8.5)/15 (0 to 20)	22.5 (8.1)/20 (20 to 20)	0.001

SD, standard deviation; IQR, interquartile range

examination confirmed the diagnosis of an osteochondroma in all patients.

Comparison of the pre- and post-operative ROM showed that the resection of the osteochondromas resulted in significant improvement of hip joint function in all directions (Table I).

The mean pain scores and RAND-36 scores are summarised in Table II. Compared with the pre-operative scores, the mean pain reduction was 4.3 (0 to 8) at rest and 5.5 (0 to 9) during activity. None of the patients reported an increase of pain post-operatively. The post-operative perception of pre- and post-operative health status showed a

**Table II.** Comparison of the pre- and post-operative pain and RAND-36 scores in 17 patients (20 hips) with multiple hereditary exostoses (MHE), and reference values for a general Dutch population with MHE, which was assessed with use of the one sample*t*-test

	Pre-operative mean (SD) median	SD) median Post-operative mean (SD) median p-valu (IQR range)	p-value	Gen MHE pop <sup>9</sup> Mean (SD)
	(IQR range)			
Pain scores (numerical rating scores)				
At rest	4.9 (2.9)/5.0 (4 to 8)	0.6 (1.5)/0.0 (0 to 1)	0.001	
During activity	7.4 (2.6)/8.0 (7 to 9)	1.9 (1.9)/1.0 (1 to 3)	< 0.001	
RAND-36				
Physical function	48.5 (23.2)/45 (30 to 57.5)	77.0 (18.5)/80 (75 to 90)	< 0.001	64.2 (25.6)
Role physical	26.3 (28.6)/25 (0 to 25)	76.3 (22.8)/75 (62.5 to 100)	< 0.001	67.7 (39.2)
Role emotional				82.4 (34.8)
Social function	67.5 (23.4)/63 (56.3 to 87.5)	83.8 (12.9)/87.5 (75 to 87.5)	0.003	80.4 (23.3)
Mental health	76.2 (17.7)/76 (62 to 95.5)	82.2 (15.3)/84 (78 to 94)	0.05	76.2 (14.5)
Pain	42.1 (19.1)/35 (35 to 51)	72.7 (16.2)/67.4 (67.4 to 72.5)	< 0.001	63.7 (24.0)
General health	54.5 (18.3)/55 (42.5 to 62.5)	60.0 (19.7)/60 (45 to 70)	0.196	66.5 (21.5)
Vitality	67.1 (18.7)/65 (52.5 to 85)	75.5 (16.5)/ 77.5 (65 to 90)	0.006	61.3 (17.6)
PCS	32.5 (8.6)/30 (27.5 to 35.2)	45.6 (6.5)/46 (42 to 47.6)	< 0.001	
MCS	54.2 (9.4)/56 (46.7 to 62.8)	55.5 (7.0)/57.3 (55.6 to 60.5)	0.872	

SD, standard deviation; PCS, physical component score, MCS, mental component score

significant improvement in seven RAND-36 sub-domains (Table II). No significant differences were found for subscales general health and mental component summary. Role limitations owing to emotional problems could not be compared because of missing data in this subscale. Preoperative RAND-36 scores of our patients were significantly lower compared with the reference RAND-36 scores of the general MHE population for sub-domains physical functioning (t-test, p = 0.007), role limitations caused by physical health problems (t-test, p < 0.001), social functioning (t-test, p = 0.024), pain (t-test, p < 0.001) and general health (t-test, p = 0.009) (Table II).

All patients were satisfied with the outcome of the operation (seven partially satisfied and ten fully satisfied).

Post-operative complications were experienced in four patients which required secondary surgical procedures. Traumatic separation of the trochanteric osteotomy because of a fall, and pseudarthrosis of the trochanteric osteotomy needed surgical intervention two and eight months, respectively, after the initial procedure. A pertrochanteric femoral fracture owing to a fall required internal fixation three months post-operatively. Another patient developed AVN of the femoral head (Ficat stage I)<sup>23</sup> requiring decompression of the femoral head 22 months postoperatively. Four years after initial surgery, he presented with recurrence of hip pain during walking and was experiencing pain at night. The MRI-scan showed progression of AVN and secondary osteoarthritis, for which he will undergo a total hip arthroplasty. Although those patients who experienced complications seemed to have less improvement in post-operative function and pain scores compared with the complication free group, the differences were not statistically significant (p-values measured with use of the one sample t-test ranged from 0.074 to 0.756 for

the five principal directions. For pain, p-values ranged from 0.173 to 0.415). No other complications such as nerve damage, wound infection or heterotopic ossification occurred.

In three patients, the screws were removed after the trochanteric osteotomy had healed because of discomfort. Post-operatively, hardware removal was performed in four asymptomatic patients in one session simultaneously with the resection of osteochondromas of other locations.

#### **Discussion**

In patients with MHE there is an estimated incidence of proximal femoral osteochondromas of approximately 80%. There are few reports on surgical removal of symptomatic lesions in the literature. Of the 61 cases reported in English literature, 24 cases involved patients with MHE 15,24-32 and 13 cases consisted of patients with a solitary osteochondroma. The remaining 24 cases were not classified. The remaining 24 cases were

The difference between the incidence of osteochondromas around the hip joint and the number of patients who are treated surgically is possibly explained by the fact that these deep-seated osteochondromas are not as symptomatic as more superficial lesions. Besides, complaints arising from osteochondromas around the hip joint may be underestimated as these symptoms are not always recognised.<sup>13</sup>

Although most of these femoral osteochondromas do cause mild pain or functional impairment, activities of daily living are usually not significantly influenced by femoral lesions. For many patients with MHE, the choice of occupation and the ability to perform sports already seems to be effected by other MHE related problems. Therefore, usually no heavy manual labour or sports are practiced by these patients. 9

Malignant transformation is the most feared complication of osteochondromas. The risk of malignant change in a solitary osteochondroma is low but the prevalence in MHE patients is significantly higher with a reported incidence of approximately 2% to 6%. 44 Clinical suspicion of malignant transformation is elicited by atypical location, large size, local pain or growth of the lesion after skeletal maturity. 3 Evaluation by MRI for the features of malignant transformation such as a mass arising from the cartilage cap and thickness of the cartilage cap > 1.5 cm to 2.0 cm in adult patients, 3 excluded neoplastic change in our series. Since centrally located osteochondromas around the hips and pelvis are more likely to undergo malignant transformation 45 patients with MHE being managed conservatively should be followed-up periodically.

Those proximal femoral osteochondromas which cause functional impairment may be considered for resection. The main surgical objectives are to obtain an adequate exposure of the lesion(s) without damaging the femoral head vascularity while protecting the neurovascular structures around the hip. The surgical approach is usually dictated by the location of the osteochondroma<sup>4,20,27,30,36</sup> each of which has some limitation or disadvantage. Whereas release of the gluteus medius and the tensor fascia lata during the anterior dislocation of the hip with the Smith-Petersen approach is associated with post-operative abductor complaints, 20 posterior approaches require release of the short external rotator muscles which may damage the MFCA and the inferior gluteal artery. 17,20 Anterolateral or lateral approaches will lead to incomplete exposure of the femoral neck and acetabulum. Arthroscopic resections have been described for small acetabular and femoral neck osteochondromas. 17,37 However, accurate contouring of the femoral neck and full access to the acetabular labrum and cartilage are difficult during hip arthroscopy.<sup>26</sup> In the currently sparse literature on multiloculated/broad based or circumferential osteochondromas, surgical approaches varied considerably from single anterior or posterior to extensive combined or the Ganz surgical dislocation of the hip. 15,24-27

Although there is no uniform strategy for treatment of proximal femoral osteochondromas, dislocation of the femoral head using the Ganz approach provides excellent exposure of the femoral neck and acetabulum in well-selected cases. This exposure facilitates complete en-bloc resection of the osteochondroma while limiting the risk of damaging intra- and peri-articular structures during resection. Furthermore, stability of the hip joint is directly restored by rigid fixation of the trochanteric osteotomy, which will allow early rehabilitation of the patient.<sup>20</sup>

Despite these advantages, AVN of the femoral head remains a concern of this surgical approach. Of the 61 previously reported cases of osteochondroma resections with various approaches, AVN was only reported once after re-operation for a recurrent osteochondroma.<sup>27</sup> Ganz et al<sup>20</sup> described use of the approach in 213 hips in patients with diagnoses mostly other than solitary or MHE-

related osteochondromas of the hip. Of these patients, none developed AVN. Despite adherence to the described technique this complication occurred in one of our patients.

Another concern of this approach is pseudarthrosis of the greater trochanter, which occurred in one of our patients. The fracture of the femoral neck that occurred in another patient three months post-operatively was not related to the approach but was probably the result of weakening of the femoral neck by resection of the osteo-chondroma.<sup>17</sup> Other complications of osteochondroma resection of the hip reported in literature such as peroneal or sciatic neuropraxia, vascular injury, heterotopic ossification and recurrent subluxation of the hip were not encountered in our patients.<sup>40,42,43</sup>

Despite the complication risk, the overall patient satisfaction rate was high and removal of the osteochondromas significantly reduced pain and increased the ROM of the hip joint.

None of the previous studies investigated the effect of the resection of osteochondromas of the hip on the health-related quality of life. MHE is often a painful disorder with profound impact on daily life. In a previous study, patients with MHE scored significantly lower on physical and social functioning, vitality, general health perception and pain in the RAND-36 compared with a random population of adults.<sup>9</sup>

Our patients had similar scores on vitality and mental health. Their pre-operative scores on physical and social functioning, limitations of activities caused by physical health problems, pain and general health were significantly lower compared with the general MHE population. For some subdomains, the pre-operative scores of our patients were even similar to quality-of-life scores (RAND-36) of patients undergoing hip arthroplasty because of end-stage osteoarthritis the serious impact of osteochondromas of the hip on quality of life.

This study presents the largest reported series of MHE patients undergoing resection of osteochondromas located on the proximal femur. We have demonstrated that resection of proximal femoral osteochondromas through a surgical dislocation of the hip according to the method of Ganz et al<sup>20</sup> can relieve symptoms with a significant decrease in pain and improvement in function and general health.

In the light of the noted risk of complications, in our opinion conservative treatment remains the first choice of treatment for mild symptoms related to osteochondromas of the hip in patients with MHE. When daily function and activities are affected by pain and limited function of the hip owing to the osteochondroma formation, resection of the lesion is warranted and can significantly improve quality of life.

#### Author contributions:

J. C. Sorel: Writing the paper, data collection, data analysis.

M. Façee Schaeffer: Writing the paper, data collection, data analysis.

A. S. Homan: Writing the paper, data collection, data analysis.

V. A. B. Scholtes: Data collection, data analysis.

D. H. R. Kempen: Writing the paper, data collection, data analysis.

S. J. Ham: Performed surgeries, writing the paper, data collection, data analysis.

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