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Post-traumatic disturbance of the epiphysis of the humeral trochlea: a spectrum of pathology

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Background: Hegemann disease and fishtail deformity are classified as growth disturbances in the physeal plate of the humeral trochlea. It is questionable if these 2 diseases should be considered as 2 distinct conditions. The aims of this study are to (1) point out similarities between both conditions, (2) discuss etiology, and (3) provide diagnostic tools.
Methods: In a multicenter prospective cohort study, 19 patients with growth disturbance of the humeral trochlea were included. Assessment consisted of a detailed history, physical examination, and standard radiographs in 2 directions. The radiographs were evaluated for skeletal age, carrying angles, and trochlear notch angles. Statistical analysis was performed using Stata.
Results: A total of 19 patients were included: 2 males (11%) and 17 females (89%). The mean age of the patients was 12.8 years. In 17 patients (89%), a traumatic injury of the elbow was reported, before presentation. Decreased trochlear notch angle (<104°) was seen in 16 patients (84%). Accelerated closure of the growth plate of the affected elbow was seen in all skeletally immature patients.
Conclusions: The main risk factor for both Hegemann disease and fishtail deformity is an injury of the elbow with open growth plates. Imaging studies support the hypothesis that both diseases are likely to be a continuum of the same process. Therefore, we propose to use 1 nomenclature for this pathologic process: post-traumatic disturbance of the epiphysis of the humeral trochlea.

Osteochondrosis is a focal disturbance in enchondral ossification within the physeal plate.[6] In general, it is believed that osteochondrosis goes through multiple stages in a fixed order, similar to Perthes disease.[4,25] Most osteochondroses are self-limiting, and symptoms will resolve by aging and modification of activities.[6]

Hegemann disease is currently known as an osteochondrosis localized at the immature humeral trochlea.[11] This is considered an idiopathic but reversible disorder of the trochlear physeal plate. It is assumed that Hegemann disease develops because of disturbances of vascular supply. However, Hegemann disease is not very well defined as is its etiology.

"Fishtail deformity" is described as a central deficiency of the epiphysis in the humerus.[28] Multiple authors have described fishtail deformity as a complication of a distal humeral fracture that occurred in childhood.[2,8,21] Still, many aspects of the etiology of both Hegemann disease and fishtail deformity remain unclear.

In adulthood, vascular supply of the medial humeral trochlea originates from the inferior ulnar collateral artery and the lateral humeral trochlea is supplied by posterior perforating vessels. The vascular supply of the lateral and medial trochlea does not anastomose (Fig. 1). This results in a hypovascular watershed area in the central and lateral part of the trochlear groove.[29] It is assumed that

Institutional review board approval from the Amphia Hospital was received for this study.

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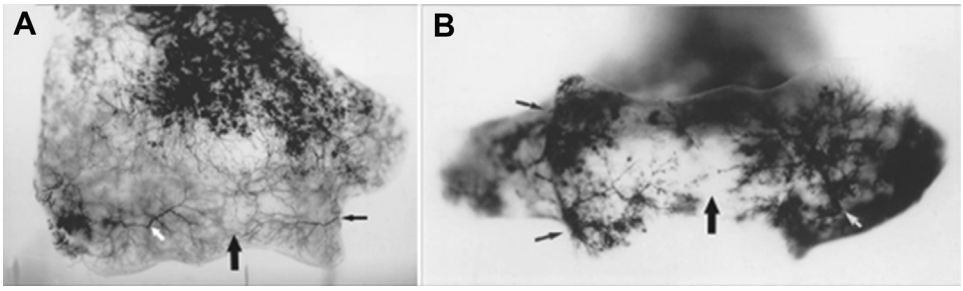

Figure 1 Photomicrograph of the blood supply of the distal humerus: Anterior (**A**) and inferior (**B**) views. The blood supply enters from the nonarticular surface via both anterior and posterior vessel (⟵). These arterioles are end arterioles and do not anastomose. The lateral aspect of the trochlea is supplied by an intraosseous vessel from the capitellum (*white arrow*), which enters posteriorly. There is a relative hypovascular area in the trochlear groove (⟵). (Copied from reference [29].)

this region is probably minimally vascularized during childhood as well.

The critical blood supply in this watershed area can be compromised because of major trauma, such as distal humeral fractures or dislocations of the elbow. Consecutive serial microtrauma caused by repeated extreme movement of the elbow as seen in some sports (eg, tennis, hockey, handball, volleyball,[12,14] and gymnastics[23]) can also result in ongoing vascular damage. Claessen et al[3] presumed that Hegemann disease and fishtail deformity might be a continuum of the same process. They suggested that Hegemann disease might be a (sometimes reversible) precursor of the fishtail deformity.

To investigate this suggestion, we (1) point out similarities between both conditions, (2) discuss etiology, and (3) provide diagnostic tools. We also propose a new nomenclature to avoid the use of different names for a continuum of the same pathologic process in the future.[23]

Methods

An institutional review board–approved observational cohort study was performed in 3 large Dutch hospitals. One of these hospitals is a large national referral center for complex elbow pathology. Data were obtained prospectively. All patients diagnosed with radiographic signs of Hegemann disease or fishtail deformity between 2008 and 2015 were included. Diagnosis was either made by plain radiographs or magnetic resonance imaging (MRI). Inclusion criteria were: (1) trochlear notch angle <104°[9] or (2) edema on MRI at the trochlear region. Exclusion criteria were signs of recent traumatic injury on plain radiographs or MRI (eg, clear fracture line without callus or edema).

At baseline, information on clinical symptoms, arm dominance, comorbidities, sports activity, medical history, and physical examination was assessed. Plain radiographs were obtained routinely. Additional imaging such as MRI or computed tomography (CT) scan was performed when clinical symptoms remained more than 3 months. The decision to perform CT or MRI was surgeon based.

Plain radiographs were assessed for skeletal maturity using the Sauvegrain method.[5,24] Trochlear notch angles and carrying angles were measured on plain radiographs using the method described by Goldfarb et al[9] (Fig. 2). These measurements were considered normal when they fitted the range of 2 standard deviations (SD) from average according to the guidelines of Goldfarb. For the carrying angle, the normal range was 6°-26° of valgus, and for the trochlear notch angle, the normal range was 104°-148°.[9]

During follow-up, plain radiographs were obtained at every visit and additional imaging was performed if clinical symptoms such as progressive pain, locking symptoms, and progressive loss of motion necessitated this. Follow-up was performed until full skeletal maturity or until symptoms completely resolved.

Statistical analysis

For statistical analysis, StataCorp Stata 15.1 (StataCorp LLC, College Station, TX, USA) was used. Variables were presented with frequencies and percentages for categorical variables and as the mean for continuous variables. To compare the Sauvegrain score in the affected elbow with the unaffected elbow, we used a Wilcoxon signed-rank test. To assess the correlation between carrying angle and trochlear notch angle of the affected side, we performed a Pearson correlation test.

Results

Patient characteristics

A total of 19 patients (2 males, 17 females) with unilateral growth disturbance of the humeral trochlea were included. The mean age was 12.8 years (range, 8-17 years). Patient characteristics are summarized in Table I.

Radiographic appearance showed no signs of elbow pathology other than growth disturbance of the trochlear physis during skeletal development. No systemic diseases were present in these patients.

Clinical presentation

The main clinical complaints of the patients (Table II) were pain (17 of 19, 89%), locking of the joint (16 of 19, 68%), and decreased range of motion (17 of 19, 89%). A history of major trauma (distal humeral fracture or elbow dislocation) was mentioned in 14 patients (74%). Nine patients (47%) participated in high-risk sports (eg, tennis, gymnastics, hockey). In the 14 patients with a history of major trauma, the exact age at trauma was known for 12 patients. The mean age at trauma for all patients was 6.5 years (SD, 3.7). The median interval between trauma and first presentation at the hospital was 75 months (range, 1-125 months).

Imaging

Measurements on the radiographs are summarized in Table III.

In 4 patients (skeletally immature at presentation), more than 3 sequential radiographs were made during follow-up. In these, early radiographs show different stages of Hegemann disease (Fig. 3, *B* and *C*),

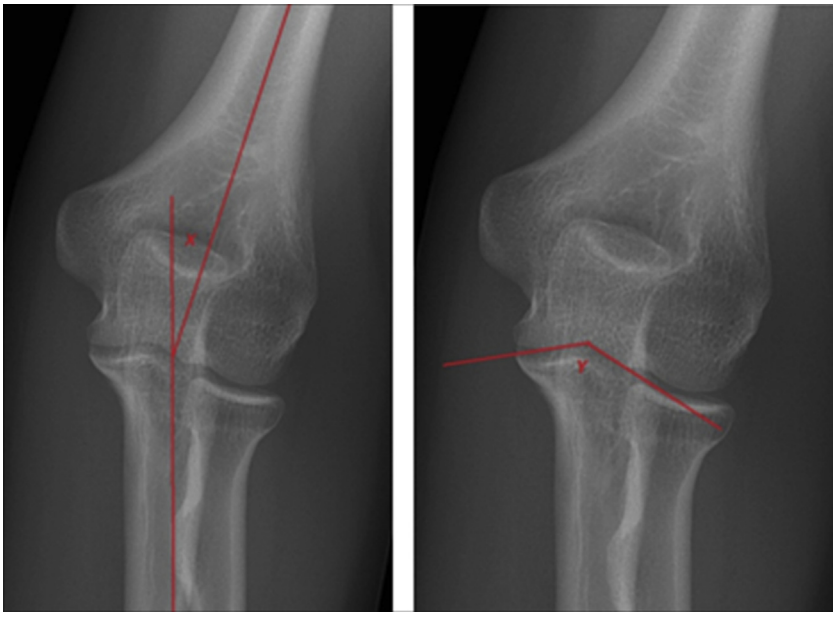

Figure 2 Method for the measurement of the carrying angle (*X*) and trochlear notch angle (*Y*).

followed by the onset of a progressive fishtail deformity on following radiographs (Fig. 3, *D*). A radiograph of the contralateral side is made for comparison (Fig. 3, *A*).

In all patients with bilateral radiographs before full skeletal maturity, the affected elbow showed advanced stages of all 4 of Sauvegrain regions, including the olecranon apophysis and the radial head, compared with the unaffected elbow. This increase was highly significant ($P = .027$, Wilcoxon signed-rank test).

No correlation between the carrying angle and trochlear notch angle of the affected side could be found (Pearson correlation coefficient: −0.018; $P = .94$).

When the clinical symptoms remained more than 3 months, an additional CT or MRI was performed. The decision to perform CT or MRI was surgeon based.

In 18 patients (95%), additional imaging was performed. CT scans were performed in 15 patients (79%), and MRI scans in 10 patients (53%).

Discussion

Both Hegemann disease and fishtail deformity are rare conditions. This can be concluded from the small number of cases in previous studies.[1,2,7,8,10,11,13,16-19,21,22,27,28] Because of the small numbers, information on incidence, etiology, and clinical outcome is limited. The lack of awareness of this condition might be related to the fact that this pathology is easily overlooked. Nevertheless, this condition can lead to severe loss of motion, pain, and locking symptoms as seen in the majority of patients in this study.

The purpose of this study was to (1) point out similarities between both conditions, (2) discuss possible etiology, and (3) provide diagnostic tools. We also propose a new nomenclature to avoid the use of different names for a continuum of the same pathologic process in the future.

In previous reports, it is suggested that Hegemann disease and fishtail deformity are a continuum of the same process of hypovascularity and secondary growth disturbances of the trochlear

Table I
Patient characteristics at baseline (n = 19).

Number	Gender	Age (yr)	Affected side	Dominant arm	Major trauma	High-risk sports
1	Female	11	Right	Right	Yes	No
2	Female	9	Left	Right	Yes	No
3	Female	17	Right	Right	Yes	Yes
4	Female	16	Right	Left	No	No
5	Female	10	Right	Right	Yes	No
6	Female	10	Left	Right	Yes	Yes
7	Female	16	Right	Right	No	Yes
8	Female	15	Right	Right	Yes	No
9	Female	11	Left	Right	No	Yes
10	Female	12	Left	Right	Yes	Yes
11	Female	11	Left	Left	Yes	No
12	Female	8	Left	Right	Yes	Yes
13	Female	14	Left	Right	No	No
14	Female	14	Right	Right	Yes	Yes
15	Female	10	Right	Right	Yes	Yes
16	Female	14	Right	Right	No	Yes
17	Male	16	Right	Right	Yes	No
18	Female	15	Right	Right	Yes	No
19	Male	15	Right	Left	Yes	No

Table II
Signs and symptoms at baseline (n = 19)

Number	Pain	Locking symptoms	Flexion <140°	Extension <0° (extension lack)	Pronation or supination <70°
1	Yes	Yes	Yes	Yes	No
2	Yes	No	No	Yes	No
3	Yes	Yes	No	Yes	No
4	No	No	Yes	Yes	No
5	Yes	Yes	Yes	Yes	No
6	Yes	No	No	Yes	No
7	Yes	Yes	Yes	No	No
8	Yes	Yes	No	Yes	No
9	Yes	No	No	Yes	Yes
10	Yes	Yes	No	Yes	No
11	Yes	No	Yes	No	No
12	No	No	Yes	Yes	Yes
13	Yes	Yes	No	Yes	Yes
14	Yes	Yes	No	Yes	No
15	Yes	Yes	Yes	Yes	Yes
16	Yes	Yes	Yes	Yes	No
17	Yes	Yes	No	No	No
18	Yes	Yes	No	No	No
19	Yes	Yes	No	Yes	No

Table III
Measurements on plain radiographs

Number	Sauvegrain score* (affected side)	Sauvegrain score* (unaffected side)	Carrying angle† (degrees of valgus)	Trochlear notch angle† (degrees)
1	26.5	26	22	52‖
2	24	20	13	90‖
3	27‡		14	88‖
4	27‡		24	92‖
5	21.5	17.5	18	86‖
6	23.5		22	103‖
7	27‡		12	77‖
8	27‡		−6§	90‖
9	16		26	109
10	24.5	19.5	8	65‖
11	23.5	20	2§	88‖
12	22.5	20	14	83‖
13	27‡		8	132
14	23.5	21	18	126
15	22.5	18	15	88‖
16	27‡	26	15	62‖
17	27‡		15	60‖
18	27‡		16	49‖
19	27‡		18	70‖

* At baseline.
† At skeletal maturity.
‡ Full skeletal maturity.
§ Outside normal range according to Goldfarb et al.[9]
‖ Decreased compared with normal range according to Goldfarb et al.[9]

physeal plate.[3] In our series, this hypothesis can be confirmed by sequential radiographic imaging in 4 patients. These sequential images illustrate the progression of Hegemann disease into a progressive fishtail deformity (trochlear notch angle <104°). The similarity in disease localization, post-traumatic onset, and clinical presentation of the 2 diseases supports this hypothesis. Therefore, we suggest the use of new nomenclature for this pathologic process: post-traumatic disturbance of the epiphysis of the humeral trochlea (PDET).

Within PDET, multiple stages can be defined (Fig. 4). Stages 1 through 4 correspond with the former stages of Hegemann disease (dispersal, condensation, calcification, regeneration) and stage 5 is an end stage (remodeling). In some, but not all, patients, stage 5 can be fishtail deformity. However, it remains unclear which patients show full recovery after stage 4 and which patients develop a fishtail deformity at stage 5. We hypothesize that damage to the trochlear physis caused by stages 1-4 can be considered reversible during remodeling, whereas malformation of the distal humerus in stage 5 is irreversible. Whether full recovery takes places depends on the amount of damage to the trochlear physis. Our hypothesis suggests that if vascular supply recovers before the closure of the physeal plate, the distal humerus will remodel into a normal trochlea at stage 5; if this does not recover, then deformity remains in stage 5. Therefore, activity modification was part of treatment in stages 1-4 of PDET. After full skeletal maturity, no progression of the deformity is expected.

Another hypothesis is that alteration to the subchondral bone of the trochlea occurs in stages 1-4 and that the damage is only irreversible if the physis gets involved in stage 5.

This series shows a high number of post-traumatic pathology (74%), and the number of post-traumatic pathology is even higher when repetitive microtrauma due to sports activities is added to the group that had a major elbow trauma (17 of 19, 89%). The high number of post-traumatic pathology seen in this series matches current literature on fishtail deformity; for Hegemann disease, this relation between the disease and trauma has not yet been

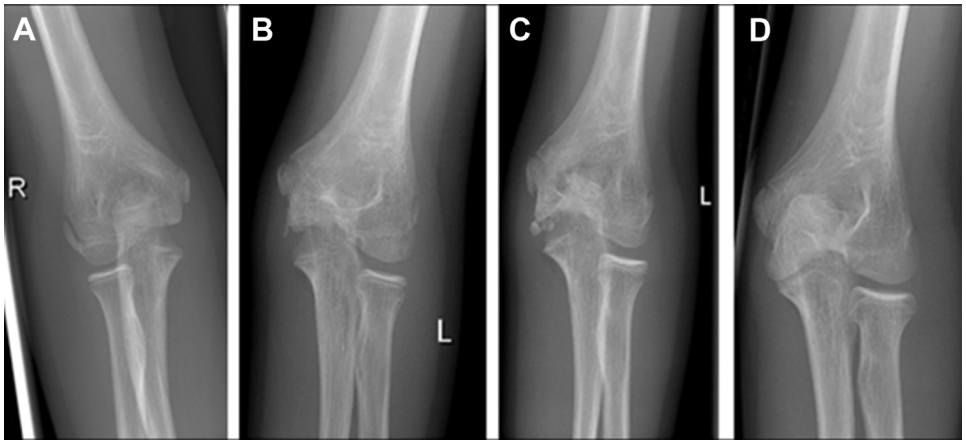

Figure 3 Sequential radiographs of 1 patient (study no. 14, female, presenting 7 months after ulnohumeral dislocation). (**A**) Unaffected side. (**B**) Baseline (age 8 years). (**C**) 1 year follow-up (age 9 years). (**D**) 4 years' follow-up (age 12 years).

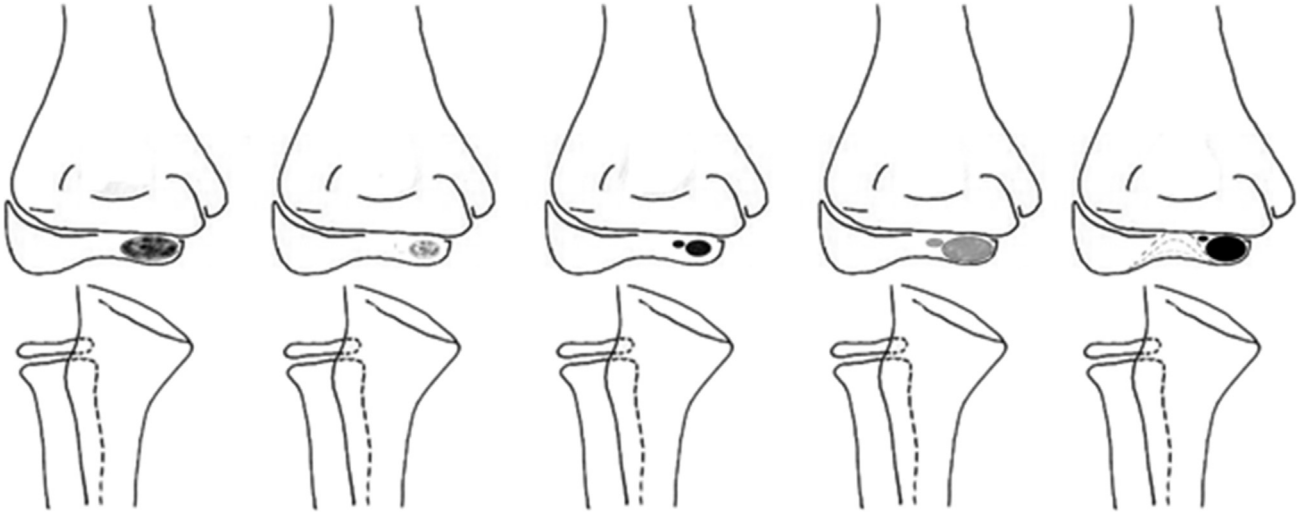

Figure 4 Stages 1-5 of post-traumatic disturbance of the epiphysis of the humeral trochlea: dispersal (1), condensation (2), calcification (3), regeneration (4), and remodeling (5).

identified. In this context, neglected trauma might be relevant, as children fall frequently. In 2017, a total of 145,700 children aged 0-14 years were treated in Dutch emergency departments for complaints due to falling.[15] Because not all falls are considered serious by a child, parent, or health care practitioner, a high number of neglected traumas remain. However, (micro)trauma of the physeal plates can still occur in these situations. In 2 patients in this series, the onset of PDET could not be related to major trauma or repetitive microtrauma. One of these 2 patients received treatment with high dose of corticosteroids for non–elbow-related causes. Corticosteroid use is known to contribute to growth disturbances in a physeal plate and avascular necrosis.[20,26] No causal factor can be identified for 1 patient in this series; this might be a case of neglected trauma.

Analysis of skeletal age using the Sauvegrain method shows an evident acceleration in skeletal maturation of the affected elbow compared with the unaffected limb. This can be partially explained by the fact that the trochlea, being the diseased region, is one of 4 regions examined in Sauvegrain score. However, accelerated maturation is not only seen in the trochlea but also in the lateral epicondyle, medial epicondyle (not included in Sauvegrain score), proximal radius, and olecranon. Affection of the complete elbow suggests that there might be other factors contributing to general growth disturbance than just local trauma to the trochlea. This is consistent with the hypothetic etiology of vascular damage in the elbow. Vascular damage is not isolated to the humeral trochlea, but vascularization of the entire elbow can be compromised by (micro) trauma, even in the absence of a fracture at that location. Also, decreased vascular supply can lead to delayed healing of microtrauma. Moreover, it is known that an increased load on the elbow can cause delayed maturation of the physeal plate. If patients have symptoms, they will probably decrease the load on the elbow, which can result in an accelerated closure of the physeal plate.

In our series, the vast majority of patients are female (89%). This is in strong contrast to numbers shown in current literature where the percentage of male patients is significantly higher.[3] Most cases were presented to us as tertiary referrals from other Dutch hospitals, so possibly some sort of bias is originated in the referring hospitals, but this remains unclear.

The measurement of trochlear notch angle seems to be the most suitable tool in diagnostics when PDET is suspected. In our series, 84% of the patients have a trochlear notch angle outside the range of ±2 SD. Unfortunately, trochlear notch angle is best measured in advanced skeletal maturity (skeletal age $\geq$12 years).[9] Before this skeletal age, the margins of the trochlea are not entirely ossified and the measurement cannot be done reliably. Therefore, trochlear notch angle is a less suitable measurement in young patients

presenting with an early stage of PDET. A more suitable measurement in the young child is not yet available and will be subject to future research.

The use of additional imaging is very common in PDET (95%). MRI is used for confirmation of diagnosis. MRI can be used for diagnosis of early stages, as well as for diagnosis in the skeletally young child. When PDET is already diagnosed, a CT scan can be used to find an(other) explanation for limited range of motion and locking, such as loose bodies or deformation. The study was most importantly limited by the small number of patients and late presentation (probably due to lack of awareness for this disease). Only 4 of our patients were seen in our hospital before the deformity was radiographically present, despite long-lasting complaints. The majority of patients already showed stage 5 of PDET at their first radiograph. Therefore, it was not possible to evaluate the radiographic development during stages 1-4 for these patients and to evaluate if preventing progression to a higher stage of this disease is possible.

Also, the inclusion of patients might be biased, as mentioned before, because inclusion was mainly done in a tertiary referral center. It is not completely clear how this bias affected the outcomes.

For advice on treatment randomization, a longer follow-up is needed. This will inevitably lead to problems due to a small number of patients available. Fig. 5 shows a treatment flowchart based on the available evidence.

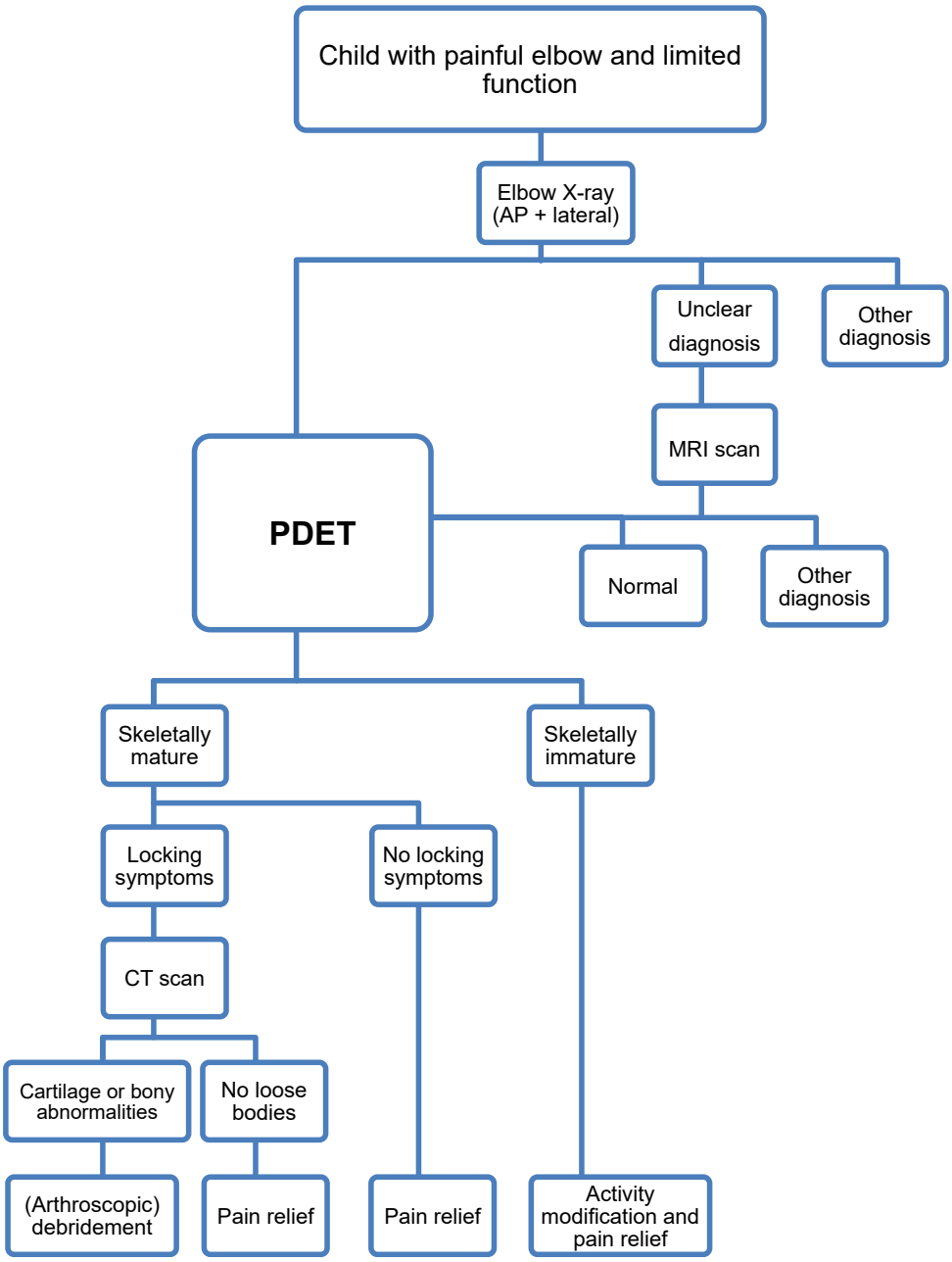

Figure 5 Treatment flowchart based on the available evidence. *PDET*, post-traumatic disturbance of the epiphysis of the humeral trochlea; *CT*, computed tomography; *MRI*, magnetic resonance imaging.

Conclusion

Our research suggests that Hegemann disease and fishtail deformity maybe a continuum of the same pathologic process. Therefore, we propose a new nomenclature: PDET. Post-traumatic vascular injury of the distal humeral physis at the vulnerable trochlear groove plays a role in etiology. The etiology is multifactorial and the exact incidence remains unclear. Pain and limited elbow function (flexion and extension) may suspect PDET. Plain radiographs with decreased trochlear notch angle confirm stage 5 of the disease. An MRI scan can diagnose earlier stages when plain radiographs are inconclusive. A CT scan is used in diagnosing bony abnormalities and loose bodies. Early diagnosis and treatment needs more attention in order to prevent end stages of PDET. Further research is initiated on incidence of PDET after distal humeral fractures and reliability of several measurements for early diagnosis of PDET. Subsequently, research on prevention and treatment of the different stages of PDET is essential.

Disclaimer

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