European Journal of Clinical and Experimental Medicine

ONLINE FIRST

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Article type: Review

Received: 13 February 2024

Accepted: 31 May 2024

Published online: 12 July 2024

eISSN: 2544-1361

Eur J Clin Exp Med

doi: 10.15584/ejcem.2024.4.4

Preiser disease in pediatric population – a review

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ABSTRACT

Introduction and aim. Preiser disease is an avascular necrosis of the scaphoid. It is a rare disease, and even less common among the pediatric population. Disease is idiopathic, occurring without trauma or scaphoid non-union. Diagnostic methods include classic radiology, CT, MRI and bone scan. Treatment may be both surgical and conservative, and main aim is pain relief and widening range of motion of the wrist. The main aim of this paper is to summarize available knowledge about Preiser disease in the pediatric population.

Material and methods. The paper is based on scientific publications available in PubMed and NCBI databases. After evaluation of abstracts, papers were selected and analyzed, considering the references citied.

Analysis of the literature. Preiser disease is rare in the pediatric population. Children tend to present better results after conservative treatment than adults. Currently there are no unified treatment recommendations in Preiser disease, as knowledge about this condition is still limited. Wider research is vital to unify management recommendations.

Conclusion. Preiser disease can affect patients of both sexes at any age, but it is exceedingly rare in children. There is a need for controlled randomized studies to establish the treatment standards, which is challenging due to the rarity of the disease. By now, non-surgical treatment tends to be the most common among the pediatric population. Surgical methods implemented in this condition include vascularized bone grafts and proximal row carpectomy.

Keywords. avascular necrosis, orthopedics, pediatric orthopedics, Preiser disease

Introduction

Preiser's disease (PD) is an idiopathic avascular necrosis (AVN) of the scaphoid. It is not connected with any trauma, acute scaphoid fracture or delayed or absent bone union.[1] The exact etiology is still undefined.[2] This disease was firstly reported by Preiser in 1910.[3,4] It is significantly less common than scaphoid fracture and Kienbock's disease, which is avascular necrosis of the lunate.[3,5] It is worth mentioning that there are reports in literature describing patients with avascular necrosis of both lunate and scaphoid, but concomitant AVN of more than one carpal bone is uncommon.[6–8] Dominant extremity is affected twice more commonly than non-dominant.[1,9] Bilateral location is extremely rare, as only a few cases were described.[2] PD has a predisposition towards females, but can affect every age group and both sexes.[4,5,10] Although, occurrence in pediatric population is exceedingly rare.[1,11] Average age of presentation is 42 years.[12]

Aim

The main aim of this review is to summarize the knowledge about Preiser disease in pediatric patients. The data is very limited due to the rarity of the disease. Review was conducted to sum up diagnostic and treatment methods in PD, but according to the literature some of the therapies were only implemented in adults and there is no data regarding its use in patients under 18 years.

Material and methods

To conduct this review, the search of PubMed and Google Scholar databases was conducted for articles raising the problem of the Preiser's disease in patients younger than 18 years. Key words such as 'preiser disease', 'idiopathic avascular necrosis scaphoid' were used. The papers were assessed based on their titles, abstracts and full texts, with the main inclusion criteria being that they described idiopathic avascular necrosis of the scaphoid in pediatric population. The most important criteria of exclusion were if the paper did not raise the problem of Preiser disease, was focused on avascular necrosis of other bones or other conditions, or pediatric cases were not mentioned. After final evaluation, 6 papers were included to the review. The review we conducted was not a systematic review.

Analysis of the literature

Typically Preiser disease occurs without prior trauma, and symptoms are not specific and lasting for months, what may lead to misdiagnosis. Manifestations include: [1,3,4,7,9,13]

- wrist pain on its dorso-radial side, including chronic pain,
- limited range of motion,
- tenderness of scaphoid area,
- swelling of dorsal side of the hand
- grip strength loss

- stiffness of the wrist.

In the literature, one can find two classifications of Preiser disease. The most popular one was proposed by Herbert and Lanzetta in 1994, consisting of 4 stages, according to radiographic scans. In 2001, Kalainov et al. suggested dividing the disease into 2 stages based on magnetic resonance images. The Kalainov classification suggest that type 1 is more likely to be connected with steroid use and worse prognosis, while type 2 tends to be linked with prior wrist trauma and prognosis is more positive. [4,9] Classifications are presented in Table 1.[3–5]

Table 1. Classifications of Preiser disease

Herbert and Lanzetta	**Kalainov et al.**
Type 1 – normal X ray, positive bone scan	Type 1 – involving the entire scaphoid
Type 2 – increased density of proximal pole	
Type 3 – proximal pole fragmentation	Type 2 – involving proximal part of scaphoid
Type 4 – carpal collapse	

According to a study conducted by Tomori and co-workers, most cases of PD concerned patients with systemic use of steroids, which is type 1 in the Kalainov classification.[7] It is connected with reducing blood circulation and bone microcirculation, caused by multiple systemic glucocorticoid intake, as they may lead to local hypertension and vascular constriction. Steroids may be also responsible for necrosis of osteocytes and osteoblasts and for increased number of bone marrow adipocytes and their hypertrophic transformation.[14] Systemic diseases reported by them that may be connected with development of post-steroid PD were renal transplantation, systemic lupus erythematosus and autoimmune hemolytic anaemia.[7,14] Although, Tomori et al. narrated a case where Preiser disease was inducted by repeated local glucocorticoid injection.[14,15] Demiroglu and colleagues described a case of PD in a five-fingered hand – in this extremely rare condition scaphoid is usually absent or hypoplastic. In their female patient bone was present, but avascular, which resulted in development of the disease.[16] In the literature one can find a case of Preiser disease development in a patient with Holt-Oram syndrome, which is a condition characterized by anomalies in the morphology of the upper limbs and cardiac abnormalities.[17,18] PD may also occur in patients with hematological diseases, such as leukemias.[19] Other factors leading to PD are presented in Table 2.[1,2,10,14,20,21] According to Konarski and co-workers, in atraumatic AVN, the use of alcohol and steroids may be responsible for osteonecrosis development in up to 80% of the cases.[21]

Table 2. Risk factors of Preiser disease

Risk factors of Preiser disease
• alcohol • nicotine • scleroderma • diabetes • chemotherapy • hemoglobinopathies • infection • vasculitis • thumb hypoplasia • cardiovascular diseases • collagen diseases

Preiser disease in pediatric population

PD is even more rare in pediatric population than among adults. This leads to the fact that treatment algorithms are unclear and not standardized.[11] In literature, 6 cases of scaphoid avascular necrosis were reported, and the youngest patient was 9 years old. Even though Preiser disease shows predisposition towards females, all pediatric cases described in literature regarded males. Risk factors were not distinguished in every case, but trauma was reported in three cases.[10,11] Three patients were treated with immobilization and the results were positive.[11] Two patients were treated surgically – one with vascularized bone graft (VBG) and proximal row carpectomy (PRC), and second one with VBG [10,11]. Summary of the cases is presented in Table 3.[10,11,22–26]

Table 3. Pediatric cases of Preiser disease reported in literature

Study	**Age/sex**	**Risk factor**	**Treatment**	**Comment**
Amundsen et al.[10]	17 M	Trauma	Pisiform vascularized bone graft and proximal row carpectomy	

Fujibuchi et al.[11]	10 M	Trauma	Immobilization for 2 months	- reported left wrist pain in pre-ossified scaphoid - ossific nucleus with fragmentation and collapse - positive results after 5 years follow-up
Jensen and Leicht[23]	10 M		Immobilization	- good clinical result was obtained
Gallie et al.[24]	12 M	Trauma	Immobilization	- no fracture was reported - MRI and bone scintigraphy revealed total ischemia of scaphoid - MRI after 6 months showed incomplete revascularization
Zheng et al.[25]	9 M		Not mentioned	- bone scintigraphy was implemented - disease did not show progression
Barthel et al.[26]	13 M	No risk factors	Vascularized bone graft	- clinical and radiological improvement was obtained

Avascular necrosis can be described as necrotic changes in the bone caused by interrupted blood supply.[21] The most common location is femoral head, but it may be present in other locations, including carpal bones.[21] Scaphoid is located on the radial side of wrist in its proximal row, and takes part in flexion, extension and radial or ulnar deflexion.[3] Morsy et al. conducted a cadaver study in 2019 which revealed that scaphoids may be divided into two types. Type I – "slender" scaphoid, tends to have wider intraosseous vascular supply. Type II – "full" scaphoid, as the vascularity is less solid, are connected with higher risk of non-union, avascular necrosis or Preiser disease.[27] Vascularization of scaphoid can be described as poor, although its AVN occurs rarely.[1] Blood supply for proximal part of scaphoid is assured by radial artery via dorsal scaphoid artery. This variant is the most common, but there are vascular variations in which blood supply is ensured by branches of radial artery and intercarpal artery. Distal scaphoid vascularization comes from radial artery or from its superficial palmar branch. Dorsal supply covers 70 to 80% of the vascularity of the scaphoid, while palmar supply covers the rest.[3,10,28]

Repetitive trauma, stress and overloading of the joint may lead to its swelling what may negatively affect intraosseus and extraosseus blood supply.[14] Interrupted blood circulation may be also caused by thrombosis,

thromboembolism or fat embolus, but the last factor is not common in carpal AVN. [29] Vascular disruption leads to ischemia, necrosis of cellular bone constituents, such as osteocytes and bone marrow, followed by bone collapse.[28,29] This process leads to painful sensations and limited range of motion and function of the wrist.[28] Among potential pathogenesis patterns of PD one can distinguish diffuse ischemic necrosis of the bone (type 1) and type 2, where avascular necrosis concerns only a part of scaphoid.[5] Although AVN are more common within carpal bones in pediatric population, one can find a case describing avascular necrosis of distal radius.[13]

When looking for diagnosis of Preiser disease one can implement classic radiology, computed tomography (CT), bone scan, arthroscopy and magnetic resonance imaging (MRI), which is considered to be a method of choice.[3,30] X- ray and CT scans are used to divide cases according to the Herbert and Lanzetta classification, which is the most widely used rating.[1,3] This classification included bone scan results as well, but nowadays this examination is replaced by MRI [10]. Bone scan with isotope typically presents enhanced ingestion in affected bone area.[31]

Classic radiology, even though is considered as initial diagnostic method, may not reveal pathological changes on early stages.[28,32,33] So when PD is suspected, magnetic resonance is suggested method, as its sensitivity is higher.[28,34] Although, bone sclerosis or fracture presence may be evaluated when analyzing X-rays. Posteroanterior and lateral views are required to make a complex diagnosis.[9] Late stages of PD may present as cystic structures, bone fragmentation or collapse on classic radiology scans.[35]

CT let physicians to visualize potential fractures and bone fragmentation or cystic structures and makes possible to assess exact morphology of the scaphoid.[9,31]

MRI is helpful when dividing cases into two types according to Kalainov classification. When osteonecrosis occurs, MRI images present signal loss in both T_1 and T_2 weighted scans. Ischemia leads to decreased signal in T_1 images, and hyperintensive images in T_2 scans. Bone marrow hyperintensivity enhanced by gadolinum administration can suggest preserved vascularity of scaphoid.[1,3,34] MRI can reveal bone edema and necrosis areas and it is helpful when assessing vascularization sufficiency of scaphoid after intravenous administration of contrast agent.[14,33,34,36,37] Type 2 in Kalainov classification – AVN concerning proximal part of scaphoid, is linked with potentially better prognosis.[30] Post-operatively, MRI scan is used to evaluate revascularization.[10]

Arthroscopy is another method that can be implemented in diagnostic process, but there is no wide report or study in this topic, and evidences are limited to case studies.[9] It allows to assess chondral surfaces and carpal ligaments, or to perform synovectomy, if needed. Arthroscopy provides a possibility not only for diagnosis, but also to treat affected wrist. In cases of fracture or bone fragmentation, curettage can be implemented.[9]

Histopathological image of PD may reveal necrotic areas with necrotic debris presence, with empty lacunae, without functional cells.[14]

There are no unified recommendations for PD treatment.[7,38] Treating AVN in pediatric patients is challenging, as it is important to avoid destruction of open physis, which may lead to limb dysfunctions.[13] Main goal in therapy is pain relief, widening range of motion and function and delaying arthritis development.[28] Early stages may be treated by immobilization, which tends to be more effective than physiotherapy, activity pattern modification or NSAIDs.[1,9,13] Hyaluronic acid injections were reported in literature as a method with good but temporary outcome.[1] It is worth remembering that these methods do not lead to slowing further PD development.[5,9,22] Surgical methods used in managing more advanced stages of PD are vascularized bone graft, proximal row carpectomy, closing radial wedge osteotomy, curettage or partial arthrodesis (capitolunate or four-corner) and 3D modelled prosthesis.[3,5,38–44] Surgical methods should be reserved for advanced cases of PD as the bone growth is not completed in children, in contrast to adults. Vascularized bone grafts may be acquisited from distal radius, iliac crest, rib or pronator quadratus pedicle.[45–49] In literature methods like scaphoid excision or its arthroscopic proximal pole resection, silicone prothesis, denervation, radial osteotomy, but according to reports, this techniques are not in a frequent use, mostly due to unsatisfactory outcomes.[3,20] Vascularized bone grafts can result in pain relief, wider range of motion and improvement of vascularization, which is assessed in MRI scan post-operatively. This method can be implemented to preserve carpal anatomy.[20]

Agarwala et al. suggested that bisphosphonate therapy may lead to the improvement in cases of avascular necrosis other than femoral head. [50] Therapies may be with alendronate only or combined – alendronate taken orally with intravenous injections of zolendronic acid. This treatment leads to collapse prevention, clinical image improvement and potential avoidance of arthroplasty. Researchers suggested that combined therapy revealed better outcomes than single alendronate inake.[50] Although, study held by Agarwala and colleagues did not include any pediatric patient to their study - youngest patients were 18 years old.

According to recent reports, the use of mesenchymal stromal cells in the treatment of osteonecrosis appears to have promising prospects, as a safe method with a rare occurrence of any complications. [51]

Although exact management recommendations for both children and adults are not established, in literature, it is suggested to initially treat young patients conservatively, especially in early stages.[1] According to Lenoir and co-workers, pediatric patients, unlike adults, may present complete cure or stop of disease progression.[22] Children affected with PD can also obtain spontaneous revascularization, in contrast to adult patients.[10,22] Researches described that both conservative and surgical approaches leaded to positive clinical outcomes.[23,24,26] It is vital to remember that AVN may occur in pre-ossified bone – case of not yet ossified scaphoid affected was described by Fujibuchi et al. Their experience show that conservative treatment with immobilization resulted in good clinical outcome.[11] Main limitation of the study was the rarity of the disease, which is even more rare in pediatric population. This leads to limited data availability. The same limitations lead to the lack of unified recommendations regarding treatment of the PD in young patients. Further studies evaluating larger groups of patients are needed to expand the knowledge about the disease,

diagnostic process and to create universal recommendations and guidelines for both surgical and conservative management methods of Preiser disease.

Conclusion

Preiser disease is a rare condition, and even less frequent in pediatric population, as literature is limited to 6 reports of patients younger than 18 years. As it is an idiopathic avascular necrosis of the scaphoid, the main symptoms include pain in dorso-radial part of the wrist and limited range of motion. Diagnostic method of choice is MRI, as classic radiology tends to appear as negative. Treatment standards are not stated, both regarding children and adults. In the pediatric population there is a predisposition to treat patients non-surgically, especially in the early stages. There is a need for more complex studies and wider research concerning this disease is needed among the pediatric population.

Declarations

Funding

Authors do not declare any source of funding.

Author contributions

The following statements should be used:

Conceptualization, K.K. and R.K.; Methodology, K.K. and R.K.; Software, K.K. and R.K.; Validation, K.K. and R.K.; Formal Analysis, K.K. and R.K.; Investigation, K.K. and R.K.; Resources, K.K. and R.K.; Data Curation, K.K. and R.K.; Writing – Original Draft Preparation, K.K. and R.K.; Writing – Review & Editing, K.K. and R.K.; Visualization, K.K. and R.K.; Supervision, K.K. and R.K.; Project Administration, K.K. and R.K.; Funding Acquisition – not applicable.

Conflicts of interest

Authors do not declare any conflict of interest.

Data availability

The data that support the findings of this study are available on request from the corresponding author.

Ethics approval

Not applicable.

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