

RESEARCH ARTICLE

The Radiological Prevalence of Incidental Kienböck Disease

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*Research performed at the Royal Infirmary of Edinburgh, The University of Edinburgh, UK**Received: 22 September 2015**Accepted: 19 November 2015***Abstract****Background:** To determine the prevalence of incidental Kienböck disease.**Methods:** A retrospective analysis of 150,912 radiological reports or images obtained over a five year period was performed of 76,174 patients who underwent a radiograph or computed tomography scan which included the wrist, in Edinburgh and Lothian, UK.**Results:** There were 5 cases of incidental Kienböck disease and 13 cases of symptomatic Kienböck disease. There were no significant differences in age, sex, ethnicity, comorbidities, smoking status, excess alcohol use or Lichtman stage between the incidental and symptomatic Kienböck groups.**Conclusion:** The radiological prevalence of incidental Kienböck disease was 0.0066% or 7 in 100,000 patients.**Keywords:** Incidental, Kienböck disease, Lunate, Prevalence**Introduction**

Robert Kienböck a Viennese radiologist described osteonecrosis of the lunate bone in 1910 (1). Multiple causes of Kienböck have been proposed, including vascular, morphological, anatomical, biomechanical, metabolic and exceptionally infective, genetic and systemic (sickle cell disease, cerebral palsy, kidney disease) etiologies are proposed for this disease, but it remains idiopathic (2). When a patient presents with wrist pain or radiological changes consistent with Kienböck disease, there is no way to know if the disease process has run its course, or if it is new and active or whether or not the disease will progress and lead to fragmentation and collapse of the lunate (3). That makes it more difficult to study whether treatments modify the course of the disease.

One measure of how often Kienböck disease arrests before it collapses is the prevalence of incidental, presumably asymptomatic and burned out Kienböck disease on radiographs obtained for other reasons. One study on the prevalence of asymptomatic, incidental Kienböck disease reported a rate of 1.9% in a South African population having radiographs at another site (e.g. leg, arm, chest) (4). In another study the incidence

of incidental Kienböck disease among middle-aged and elderly Japanese women was 1.2% (5). A study of 1450 cadavers discovered four lunate bones with pathological fracture lines at a rate of 0.28% (6).

This study determined the incidental prevalence of Kienböck disease in a British population, in and around Edinburgh, of patients having radiographs and computed tomography scans for other reasons. We also tested the null hypothesis that there are no factors associated with incidental and symptomatic Kienböck disease on hand, wrist or forearm radiographs or computed tomography (CT) scans.

Materials and Methods**Study design, setting and patient selection**

This retrospective cohort study was considered to be an audit and therefore did not require formal ethical approval with a waiver of informed consent granted in accordance with local guidelines. All adult patients aged 18 years or greater with CT scans and radiographs of hand, wrist or forearm that included the carpus between July 2008 and October 2013 in the Royal Infirmary of Edinburgh and St John's Hospital were included. Incidental Kienböck disease was defined as patients who

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were not seeking care for wrist pain due to Kienböck disease but had radiological changes consistent with Kienböck disease. Symptomatic Kienböck disease was defined as patients seeking care for wrist pain due to Kienböck disease and with radiological changes consistent with Kienböck disease. There were 76,174 patients with a total of 150,912 radiology reports (150,231 radiographs and 681 CT scans).

Outcome measures and explanatory variables

The primary outcome measure was the prevalence of incidental Kienböck disease. Search fields with the terms "Kienböck" and "lunate" were applied to identify all reports with potential Kienböck disease. All returned reports with an abnormal description of the lunate were then individually reviewed to diagnose Kienböck disease using radiographic diagnostic criteria: linear fracture lines, lunate sclerosis, collapse of lunate on radial border, changes in carpal alignment and height, fixed scaphoid rotation, proximal migration of capitate and severe lunate collapse with intra-articular degeneration of midcarpal or radiocarpal joint. The principal investigator reviewed cases where there was diagnostic uncertainty.

Uncertain cases with other potential causes for radiographic abnormalities of the lunate (n=11) were reviewed and classified as negative for Kienböck disease. This included lunate subluxation or dislocation after acute trauma (n= 6) and developmental or congenital lunate triquetral coalition (n=5).

Medical notes of patients with a diagnosis of Kienböck

disease were reviewed. The incidental group comprised patients without a prior diagnosis of Kienböck disease and an absence of wrist symptoms including pain, swelling, weakness and restriction of range of movement. The assessment of factors associated with Kienböck disease considered the following explanatory variables: age of patient during imaging, sex, ethnicity, specific medical comorbidities (chronic pulmonary disease, heart failure, malignancies, peripheral vascular disease, cerebrovascular disease, diabetes, liver disease, dementia, obesity, neurological disorders and renal diseases), smoking status, and excess alcohol intake (defined as consumption of units above current national guidelines of 21 units for men and 14 units for women).

Statistical analyses

The prevalence of incidental, symptomatic and overall Kienböck disease is presented as a percentage of the total number of patients in this study. Categorical variables are summarized as frequencies and percentages. Continuous variables are presented as mean \pm confidence intervals. The demographic and clinical characteristics of patients in the incidental and symptomatic Kienböck groups were evaluated using bivariate analysis. The chi square test was performed to calculate *p* values for categorical variables or two-sided Fisher's exact test in case the minimum expected cell frequency was less than five and the Student *t* test for continuous variables. A *P* value < 0.05 was considered significant.

Table 1. Bivariate analysis of factors associated with Kienböck disease

	Incidental Kienböck (n=5)	Symptomatic Kienböck (n=13)	P Value**	All Kienböck (n=18)
	Mean (95% CI*)	Mean (95% CI)		Mean (95% CI)
Age in years	43 (21-66)	41 (31-51)	0.77	42 (34-50)
	n (%)	n (%)		n (%)
Men	3 (60%)	11 (85%)	0.53	14 (78%)
Ethnicity				
White British	5 (100%)	10 (77%)		15 (83%)
European	0 (0%)	1 (8%)	0.99	1 (6%)
African/South American	0 (0%)	1 (8%)		1 (6%)
Asian	0 (0%)	1 (8%)		1 (6%)
Imaging Modality				
Radiograph	4 (80%)	7 (54%)	0.60	11 (68%)
Computed Tomography	1 (20%)	6 (46%)		7 (32%)
Medical Co-morbidities				
No co-morbidities	3 (60%)	5 (39%)	0.45	8 (44%)
1 - 3 co-morbidities	1 (20%)	7 (54%)		8 (44%)
>3 co-morbidities	1 (20%)	1 (8%)		2 (11)
Smoking Status				
Smoker	2 (40%)	9 (69%)	0.33	11 (61%)
Non-smoker	3 (60%)	4 (31%)		7 (39%)
Alcohol Intake				
Below limits	4 (80%)	8 (62%)	0.62	12 (67%)
Above limits	1 (20%)	5 (39%)		6 (33%)

* CI: confidence interval

** Compared between Incidental and Symptomatic Kienböck groups

Table 2. Incidental, Symptomatic and All Kienböck disease per Lichtman stage (n=18)

	Incidental Kienböck	Symptomatic Kienböck	P-Value	All Kienböck
Lichtman stage	n (%)	n (%)		n (%)
Stage I	0 (0.00%)	2 (15%)		2 (11%)
Stage II	3 (60%)	5 (39%)		8 (44%)
Stage III	0 (0.00%)	3 (23%)	0.71	3 (17%)
Stage IV	2 (40%)	3 (23%)		5 (28%)
Total	100%	100%		100.0%

Results

There were 18 wrists with Kienböck disease in 150,912 radiological examinations of 76,174 patients. Thirteen/eighteen (72%) of these patients were seeking care for wrist pain due to Kienböck disease and the diagnosis was an incidental finding in five/eighteen (28%) patients. The radiological prevalence of incidental Kienböck disease was 0.0066% or 7 in 100,000 patients. With the number of radiographs available, there were no significant differences in age, sex, ethnicity, comorbidities, smoking status, and excess alcohol use. [Table 1] There was also no difference in Lichtman stage between the incidental and symptomatic Kienböck groups ($P=0.71$) [Table 2].

Discussion

On initial diagnosis of Kienböck disease, there is currently no way to know if the disease will progress and merits potentially disease modifying treatment. If the disease has already stopped progressing then surgery to attempt to modify the course of the disease is unhelpful. The prevalence of incidental, presumably asymptomatic and burned out Kienböck disease on radiographs obtained for other reasons might give us some idea of how often the disease arrests prior to complete collapse of the lunate. This study reviewed five years' imaging of the wrist in two institutions (The Royal Infirmary of Edinburgh and St John's Hospital) to determine the prevalence of incidental Kienböck disease.

The study has a number of limitations that might have made the estimated prevalence of incidental Kienböck lower than the true prevalence. We relied on complete and accurate documentation in radiology reports. There were also search strategy limitations: potentially misspelt permutations of the terms "Kienböck" and "lunate" (with stem words spelling errors: Kien-, Kein-, -boch and variations of reference to the lunate: lunatomalacia, osteonecrosis, avascular necrosis) could not be entered into the search fields of the electronic radiological database. The majority of images were radiographs, which are not a sensitive test for Lichtman stage I Kienböck disease. The strength of this study is its large sample size and availability of a complete imaging database comprising radiographs and computed tomography scans

The prevalence of incidental Kienböck disease was approximately 1 in 15,000 patients (0.0066%) in our study. That is much lower than the prevalence in South Africa (1.9%) and Japan (1.2%), and also lower than in German cadavers (0.28%). Our study might be more accurate as it investigated a large sample number. Also it was not subject

to selection bias from specific subpopulations as in the previously reported studies. A study into elderly patients showed Kienböck to increase with age, so the prevalence in the study of elderly Japanese might be expected to be higher; however they did not include males (5, 7). In our study, it is possible that we missed a number of grade 1 early cases that were not seen or reported on radiographs. Also we may have missed cases that were reported, but the key terms were misspelled. Future studies might look at the prevalence of incidental Kienböck on MRI, but it's unusual to order an MRI in the absence of wrist problems.

This study did not identify any predisposing risk factors associated with Kienböck disease or a correlation between Lichtman stage and symptoms. Palmer reported poor correlation findings between Kienböck disease and its hypothesized etiology which include anatomic, vascular and mechanical factors (8). Anatomy of blood vessels were initially thought to contribute to the disease as classically only one single volar or one single dorsal vessel supply to the lunate had been found in some specimens (9). However this is now thought to have little influence on the development of Kienböck disease after consistent dorsal and palmar nutrient arteries were both demonstrated in larger anatomical studies (10). A meta-analysis studying ulnar variance as a potential predisposing mechanical factor, where the lunate is hypothesized to be under uneven axial force distribution from the radial aspect due to a short ulna, did not reveal any significant association (11). Mirabello et al. also describe a lack of correlation between function and Lichtman class (12).

Long-term follow-up of a large cohort of patients with Kienböck disease that chose not to have surgery would help clarify the natural history of incidental Kienböck disease. This would help establish the proportion of incidental cases that may flare and become active disease or remain arrested and burned out before complete collapse. If it turns out that a high percentage of Kienböck disease does not progress, then patients could be better informed and consider nonspecific treatment and monitoring.

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