



## Osteochondritis dissecans of the humeral capitellum in identical twin baseball players



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Osteochondritis dissecans (OCD) is a focal idiopathic alteration of subchondral bone; it has an inherent risk of instability and disruption of adjacent articular cartilage that may result in premature osteoarthritis.<sup>7</sup> Occurrence of OCD is common in the elbow and knee joints. OCD of the humeral capitellum generally affects adolescent athletes (eg, baseball players and gymnasts) who engage in repetitive overhead or upper extremity weight-bearing activities.<sup>3,5</sup> The prevalence of capitellar OCD in baseball players ranging from 10 to 12 years of age has been reported to be 2.1/1,000.<sup>12</sup> The injury can progress to the separation of a segment of cartilage and subchondral bone, forming a loose body.<sup>1</sup>

Many etiological theories have been proposed for the onset and progression of OCD, including trauma, diet, rapid growth, anatomic characteristics, lack of blood supply, necrosis of subchondral bone, and heredity.<sup>2,6,16,20</sup> The exact etiology of OCD of the humeral capitellum is still unclear, but the primary cause is believed to be repetitive trauma to the poorly vascularized capitellum.<sup>9</sup> Genetic factors are also thought to play a role in the pathogenesis; however, the genetic basis of OCD remains unknown.<sup>2</sup> To our knowledge, the unique presentation of OCD affecting male identical twin baseball players, in the absence of syndromic features, has not been reported before. Here, we present 11-year-old identical twin baseball players with almost

identical radiographic, computerized tomography, magnetic resonance imaging, ultrasound, and macroscopic findings and clinical course of OCD of the humeral capitellum.

### Case report

The patients were 11-year-old male identical twins who had been actively engaged in baseball for 4 years. The patients practiced baseball four times per week and were right-handed. The clinical history was unremarkable with no medical records or history of previous major trauma to the elbow. They were of similar physique, 147 cm tall, weighing 35 kg, and developmentally normal for their age. Twin A played mainly in the short-stop position. Twin B played mainly in the catcher position. Informed consent was received to report this case.

The twins participated in a baseball elbow medical examination held at the beginning of the off-season and were diagnosed with OCD of the humeral capitellum by ultrasonography. Three months before the examination, both twins had experienced elbow pain on throwing during baseball but had no pain on throwing at the time of the examination. During the examination, the range of motion, valgus stress test, and tenderness of the medial epicondyle or humeral capitellum were assessed physically. Twin A had a slight limitation in the range of motion in elbow extension and flexion. Twin B did not indicate any abnormalities in the physical findings. Thereafter, ultrasonography of the medial epicondyle and humeral capitellum revealed similar OCD findings (Fig. 1) and medial epicondyle abnormalities in both twins, who were advised to visit an orthopedic hospital.

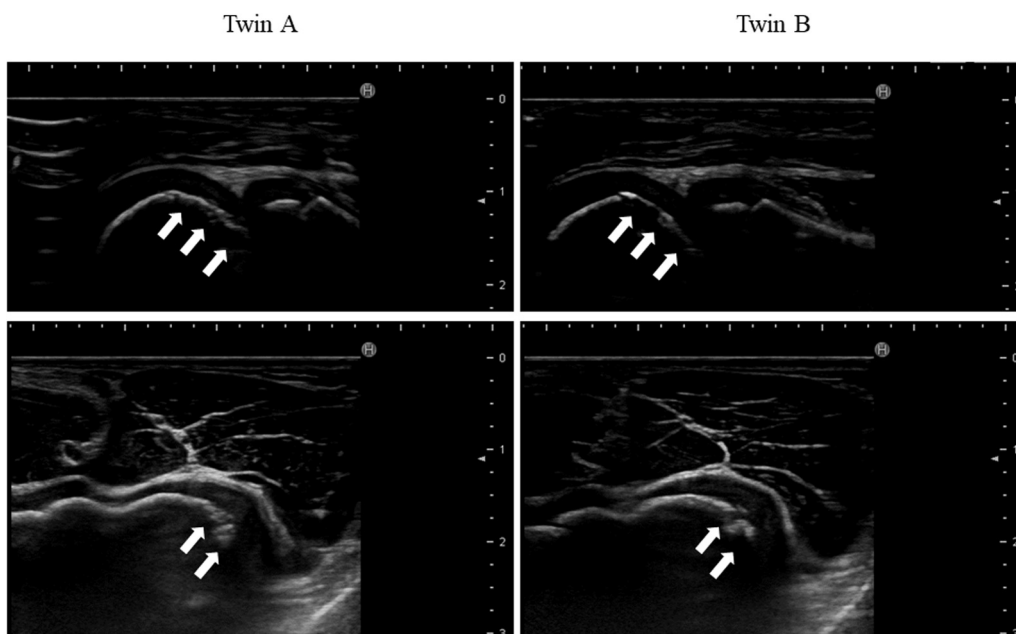
Approval for this study was received from the Ethics Committee of the Kanazawa University Hospital (approval no. 1924). Each author certifies that all investigations were conducted in accordance with the ethical principles of research.

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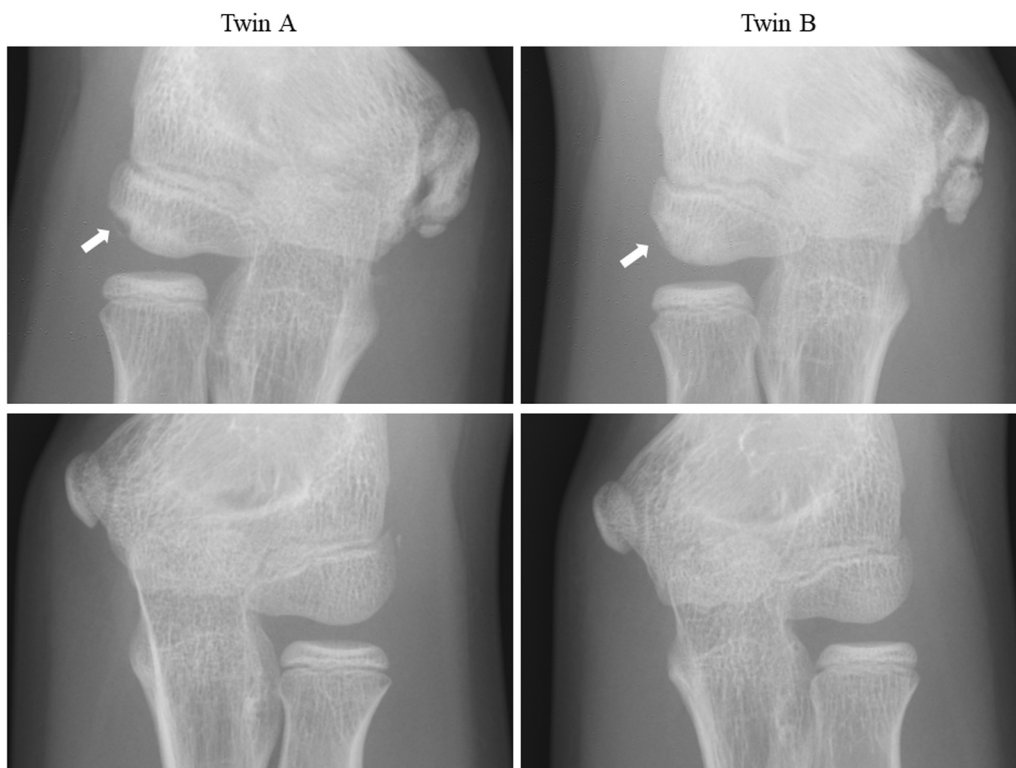
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**Figure 1** Ultrasound images at the time of baseball elbow medical examination. Images in the *top* row are a sagittal section, and those in the *bottom* are a coronal section. Almost the same subchondral bone irregularity can be seen at almost the same location at the humeral capitellum (*white arrows*).



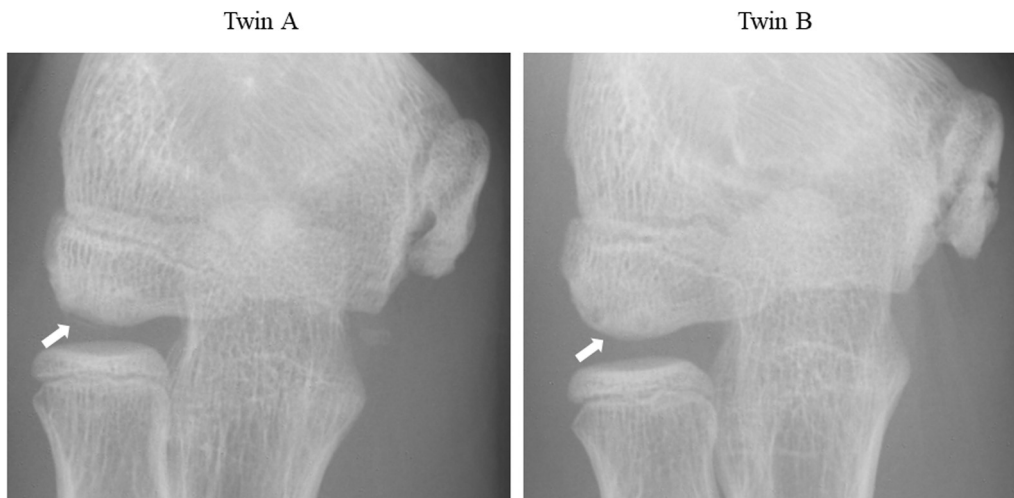
**Figure 2** Radiographs at the first hospital visit. *Top* images are of the right elbow; the *bottom* images are of the left elbow. Forty-five-degree tangential view imaging shows OCD lesions in the lucent stage at almost the same location (*white arrows*). No lesions were found on the healthy side.

One month later at the orthopedic hospital, X-ray examination revealed almost the same OCD findings (Fig. 2). The twins were instructed to cease throwing and to visit a doctor every other month.

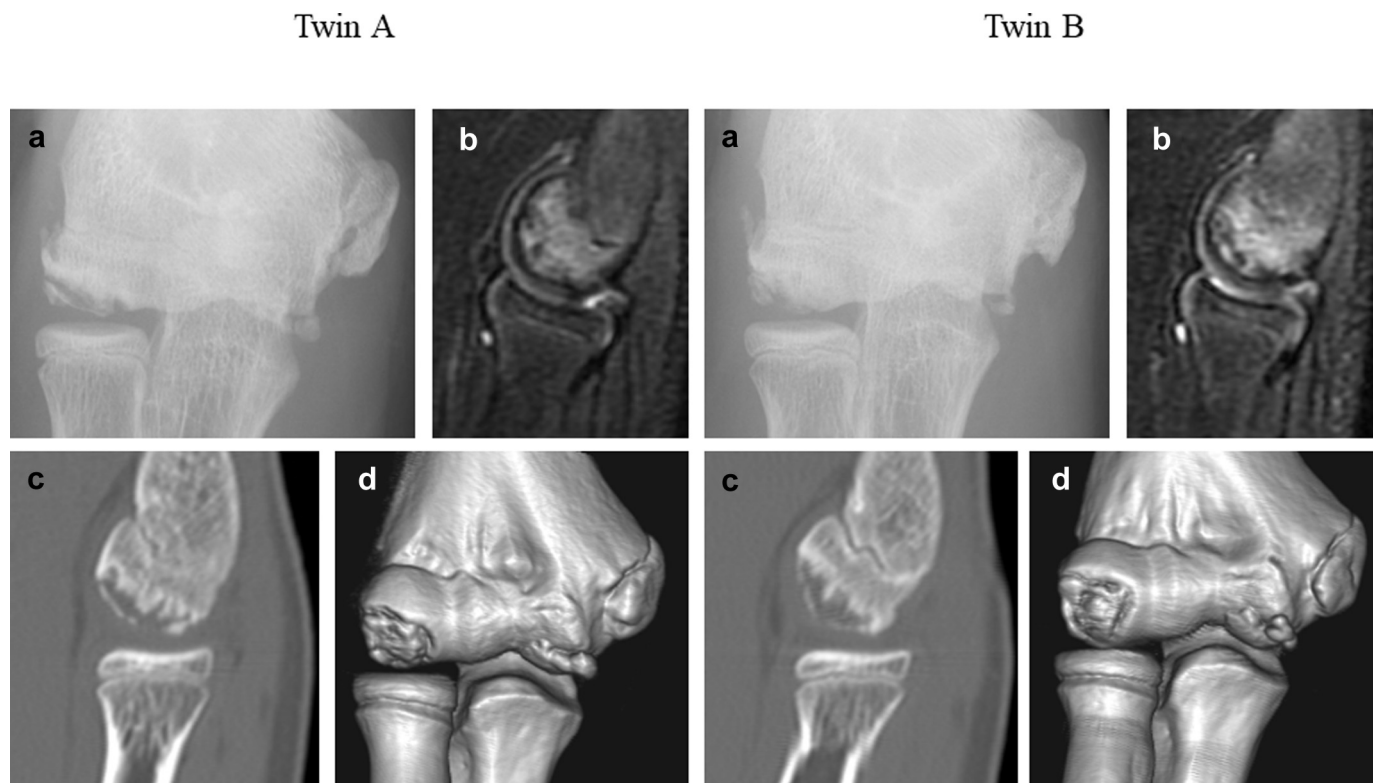
Two months later, the patients were gradually allowed to recommence throwing. After undergoing an X-ray examination 6

months after the first visit (Fig. 3), the patients stopped visiting the hospital.

After the last hospital visit, the twins completed the entire baseball season (lasting for six months) without pain in their elbows. Twin A played mainly in the pitcher position, and twin B in the catcher position during the last season. However, when they



**Figure 3** Radiographs after 6 months from first hospital visit. Lucent area extends to the center of the capitellum.



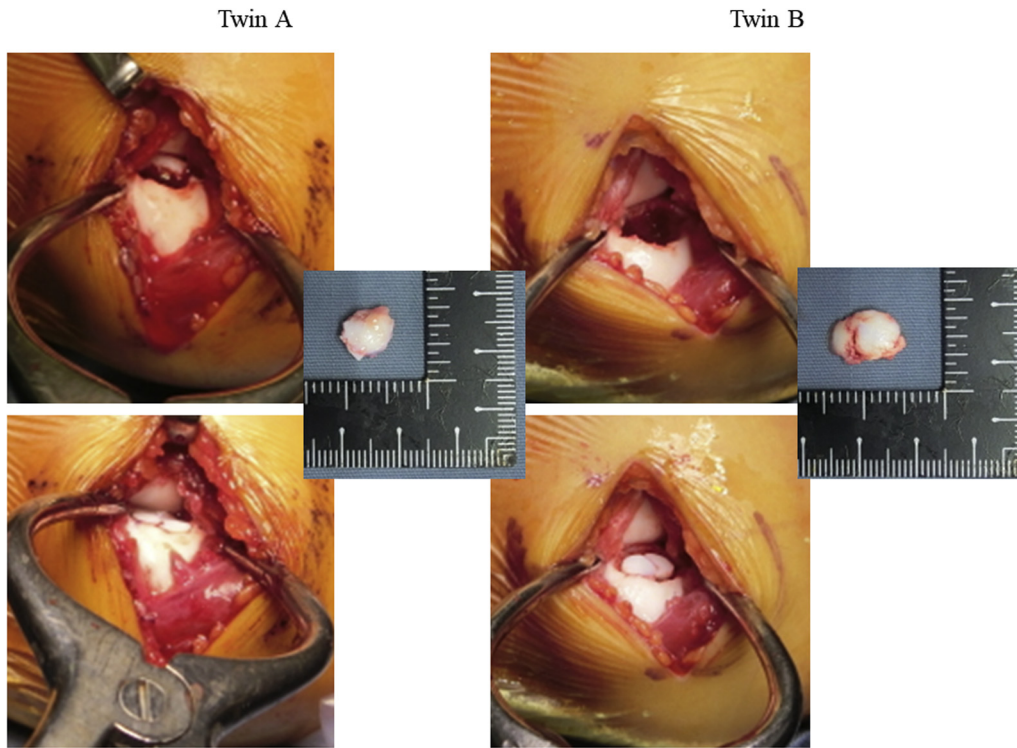
**Figure 4** Image findings after the season (1 year after the first visit). (a) The lesion is enlarged, and a fragmentation can be seen on radiographs. (b) Short tau inversion recovery (STIR) magnetic resonance imaging of the humeral capitellum showed signal changes around the OCD lesion. (c) A large area of bone-lucent images can be seen on the sagittal CT images. (d) Large lesions of almost same shape can be seen on 3DCT images.

returned to the orthopedic hospital after the season, they indicated that the OCD had worsened (Fig. 4). They hoped to continue playing baseball and decided to undergo surgery after discussion with their parents.

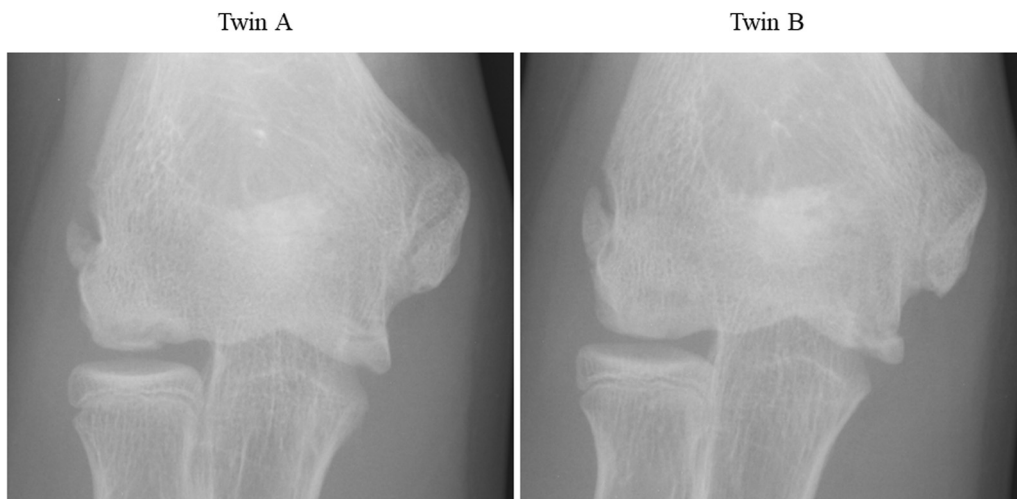
They underwent surgery to transplant the auto osteochondral graft from the unloaded part of the knee joint into the OCD lesion. Twin A had a 9 × 10-mm osteochondral fragment removed, and φ4.5-mm and φ3.5-mm osteochondral grafts were transplanted.

Twin B had a 9 × 13-mm osteochondral fragment removed, and two φ4.5-mm osteochondral grafts were transplanted (Fig. 5).

A splint was fixed for 2 days after the surgery, and then the range of motion for training was initiated. Two and a half months after the surgery, throwing exercises were gradually started. Four months after the surgery, they were allowed to throw at maximal effort and returned to playing competitively. The twins were able to play baseball without any pain when throwing; there were no



**Figure 5** Intraoperative photographs. In both patients, free cartilage fragments were removed, and osteochondral grafts were transplanted.



**Figure 6** Radiographs at the six months post-surgery. The lucent area remained in twin A, but the repair was good in both patients.

abnormal physical findings when they visited the hospital six months after the surgery (Fig. 6).

**Discussion**

We have documented our experience with 11-year-old identical twin baseball players with almost identical radiographic, computerized tomography, magnetic resonance imaging, ultrasound, and macroscopic findings and clinical course of OCD of the humeral capitellum. OCD of the humeral capitellum has been well described in the dominant arm of Little League baseball pitchers and is thought to be caused by repetitive increased loading of the radio-capitellar joint.<sup>4</sup> This experience with OCD cases of identical twin

baseball players suggests that heredity is also an important factor for OCD.

The exact etiology of OCD of the capitellum is presently unknown, but repetitive trauma to the poorly vascularized capitellum is believed to be the primary cause.<sup>9</sup> It is postulated that the mechanics during pitching places increased valgus stress and compression on the elbow, particularly during late cocking and early acceleration.<sup>5</sup> Differences in biochemical properties of the radial head and capitellum are also believed to contribute to microtrauma, with the stiffer radial head articulating with the softer lateral capitellar surface causing increased strain.<sup>18</sup> McManama et al reported repetitive trauma from throwing sports as the etiology in half of 14 patients who underwent surgical

management for capitellar OCD lesions; however, it remains unclear as to why the other half had OCD lesions.<sup>14</sup> Therefore, it seems that repetitive microtrauma is not the only cause of OCD.

Ischemia has also been implicated as a potential contributor to the progression of OCD because of its vascular anatomy. Haraldsson et al noted that the predominant blood supply was from endarteries entering posteriorly and passing through compressible epiphyseal cartilage, with no connection to adjacent metaphyseal vessels.<sup>9</sup> This tenuous blood supply is at risk of repetitive elbow injury.

It is generally accepted that repetitive microtrauma and ischemia are the leading causes of OCD lesions in the capitellum; however, there have been sporadic reports of a genetic link. To date, this predisposition for OCD in certain families has not been linked to any specific gene. Familial OCD is a disease in which articular cartilage on the surface of the knee, ankle, and elbow joints separates from the subchondral bone tissue. Stattin et al reported that this disease is caused by a missense mutation in the *agrecan* gene, which is a typical cartilage component.<sup>19</sup> The identical twin baseball player OCD cases suggest that heredity is one of the most important factors. Although genetic testing was not performed in this case, genetic factors are expected to be elucidated with the development of genetic testing in the future.

Several cases of OCD of the humeral capitellum in twins have been reported previously. OCD of the nondominant elbow in fraternal twins was reported by Kenniston et al.<sup>10</sup> Matsuura et al reported cases of fraternal twins who had been actively engaged in competitive rhythmic gymnastics with similar bilateral OCD of the capitellum.<sup>13</sup> Pudas et al reported cases of identical twin ice hockey players.<sup>17</sup> The twin cases in this study showed very similar image findings and clinical courses compared to those of the twin OCD cases reported in the literature.<sup>10,13,17</sup> Genetic factors may account for a large proportion of several potential factors for OCD.

Screening using ultrasonography enables early detection and provides an opportunity for successful conservative treatment.<sup>11</sup> Harada et al reported that ultrasonography was useful and reliable (as confirmed by radiographic findings) for detecting elbow injuries.<sup>8</sup> Ultrasonography is very useful for routine examination of OCD because it is noninvasive and cost-effective.<sup>15</sup> Ultrasonographic imaging has been used to diagnose early OCD without elbow pain in baseball elbow medical examinations.<sup>11</sup> Our cases also showed asymptomatic OCD on ultrasonography and were treated conservatively. Unfortunately, continuing baseball after conservative treatment exacerbated the condition of OCD, and surgical treatment was required. However, the period of pitching restrictions was off-season, and it was possible to return to competitive sport four months after the surgery. Early detection of OCD by ultrasonography may have had a positive effect on minimizing the withdrawal period in these cases.

## Conclusion

We have described the OCD of the capitellum in identical twin baseball players. As these were identical lesions in identical twins, this supports the theory that OCD has a genetic background. For OCD, screening by ultrasonography is considered effective for early detection and provides an opportunity for early treatment.

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**Consent:** The patients provided informed consent.

## References

1. Barrie HJ. Osteochondritis dissecans 1887–1987. A centennial look at König's memorable phrase. *J Bone Joint Surg Br* 1987;69:693–5.
2. Bates JT, Jacobs JC Jr, Shea KG, Oxford JT. Emerging genetic basis of osteochondritis dissecans. *Clin Sports Med* 2014;33:199–220. <https://doi.org/10.1016/j.csm.2013.11.004>.
3. Bradley JP, Petrie RS. Osteochondritis dissecans of the humeral capitellum. Diagnosis and treatment. *Clin Sports Med* 2001;20:565–90.
4. Brown R, Blazina ME, Kerlan RK, Carter VS, Jobe FW, Carlson GJ. Osteochondritis of the capitellum. *J Sports Med* 1974;2:27–46.
5. Cain EL Jr, Dugas JR, Wolf RS, Andrews JR. Elbow injuries in throwing athletes: a current concepts review. *Am J Sports Med* 2003;31:621–35. <https://doi.org/10.1177/03635465030310042601>.
6. Edmonds EW, Polousky J. A review of knowledge in osteochondritis dissecans: 123 years of minimal evolution from König to the ROCK study group. *Clin Orthop Relat Res* 2013;471:1118–26. <https://doi.org/10.1007/s11999-012-2290-y>.
7. Edmonds EW, Shea KG. Osteochondritis dissecans: editorial comment. *Clin Orthop Relat Res* 2013;471:1105–6. <https://doi.org/10.1007/s11999-013-2837-6>.
8. Harada M, Takahara M, Sasaki J, Mura N, Ito T, Ogino T. Using sonography for the early detection of elbow injuries among young baseball players. *Am J Roentgenol* 2006;187:1436–41. <https://doi.org/10.2214/AJR.05.1086>.
9. Haraldsson S. On osteochondrosis deformans juvenilis capituli humeri including investigation of intra-osseous vasculature in distal humerus. *Acta Orthop Scand Suppl* 1959;38:1–232.
10. Kenniston JA, Beredjickian PK, Bozentka DJ. Osteochondritis dissecans of the capitellum in fraternal twins: case report. *J Hand Surg Am* 2008;33:1380–3. <https://doi.org/10.1016/j.jhssa.2008.05.008>.
11. Matsuura T, Iwame T, Iwase J, Sairoyo K. Osteochondritis dissecans of the capitellum: review of the literature. *J Med Invest* 2020;67:217–21. <https://doi.org/10.2152/jmi.67.217>.
12. Matsuura T, Suzue N, Iwame T, Nishio S, Sairoyo K. Prevalence of osteochondritis dissecans of the capitellum in young baseball players: results based on ultrasonographic findings. *Orthop J Sports Med* 2014;2:2325967114545298. <https://doi.org/10.1177/2325967114545298>.
13. Matsuura T, Wada K, Suzue N, Iwame T, Fukuta S, Sairoyo K. Bilateral osteochondritis dissecans of the capitellum in fraternal twins: a case report. *JBJS Case Connect* 2017;7:e44. <https://doi.org/10.2152/jmi.67.217>.
14. McManama GB Jr, Micheli LJ, Berry MV, Sohn RS. The surgical treatment of osteochondritis of the capitellum. *Am J Sports Med* 1985;13:11–21.
15. Parker L, Nazarian LN, Carrino JA, Morrison WB, Grimaldi G, Frangos AJ, et al. Musculoskeletal imaging: medicare use, costs, and potential for cost substitution. *J Am Coll Radiol* 2008;5:182–8. <https://doi.org/10.1016/j.jacr.2007.07.016>.
16. Polousky JD. Juvenile osteochondritis dissecans. *Sports Med Arthrosc Rev* 2011;19:56–63. <https://doi.org/10.1097/JSA.0b013e31820b94b9>.
17. Pudas T, Koskinen SK, Hiltunen A, Mattila KT. Osteochondritis dissecans of the humeral capitellum in identical twins. *Acta Radiol Short Rep* 2012;1:arsr.2012.120004. <https://doi.org/10.1258/arsr.2012.120004>.
18. Schenck RC Jr, Athanasiou KA, Constantinides G, Gomez E. A biomechanical analysis of articular cartilage of the human elbow and a potential relationship to osteochondritis dissecans. *Clin Orthop Relat Res* 1994;305–12.
19. Stattin EL, Wiklund F, Lindblom K, Onnerfjord P, Jonsson BA, Tegner Y, et al. A missense mutation in the *agrecan* C-type lectin domain disrupts extracellular matrix interactions and causes dominant familial osteochondritis dissecans. *Am J Hum Genet* 2010;12:126–37. <https://doi.org/10.1016/j.ajhg.2009.12.018>.
20. Ytrehus B, Carlson CS, Ekman S. Etiology and pathogenesis of osteochondrosis. *Vet Pathol* 2007;44:429–48. <https://doi.org/10.1354/vp.44-4-429>.