JNMS Journal of Nippon Medical School https://www.nms.ac.jp/sh/jnms/

Case Report

Intraosseous Pneumatocyst of the Scapula: Report of Two Cases

Yasuyuki Kitagawa[1], Kazuma Miura[1], Daisuke Fukuhara[1,2], Naoto Kotani[1], Shoko Sasaki[1], Yosuke Shinozuka[1], Ryu Tsunoda[1] and Tokifumi Majima[3]

[1]Department of Orthopaedic Surgery, Nippon Medical School, Tama Nagayama Hospital, Tokyo, Japan
[2]Department of Orthopaedics, Graduate School of Medical Science, Kyoto Prefectural University of Medicine, Kyoto, Japan
[3]Department of Orthopaedic Surgery, Nippon Medical School, Tokyo, Japan

An intraosseous pneumatocyst is a gas-containing cystic bone lesion that is not associated with infection or bone necrosis due to vertebral compression fracture. The most common sites are the pelvis and spine, but it is reported rarely in other sites. To our knowledge, only two cases of intraosseous pneumatocyst in the scapula have been reported. Herein, we report two cases of intraosseous pneumatocyst of the scapula, in a 41-year-old woman and a 51-year-old man. Neither patient had shoulder symptoms, and the lesions were found incidentally during imaging studies. Plain X-rays revealed cystic lesions with sclerotic rims, located from the scapular neck to the glenoid, adjacent to the shoulder joint. Plain X-rays of the 51-year-old male patient showed osteoarthritis of the shoulder, including joint space narrowing and osteophyte formation. CT was used for diagnosis in both cases. The patients remained pain-free throughout the follow-up period (10 years and 6 months, respectively). Their lesion sizes were unchanged, and radiolucency was reduced at the final follow-up. The shoulder joint has the largest range of motion in the human body; thus, the vacuum phenomenon may occur when the shoulder is elevated or externally rotated. We speculate that gas was produced in an intraosseous ganglion or subchondral cyst, the likely pre-existing lesions, after the vacuum phenomenon occurred in the shoulder joints of these patients.
(J Nippon Med Sch 2026; 93 (2): 201–206. https://doi.org/10.1272/jnms.JNMS.2026_93-202)

Keywords: intraosseous pneumatocyst, scapula, shoulder, vacuum phenomenon, ganglion

Introduction

An intraosseous pneumatocyst (IP) is a gas-containing cystic bone lesion that is not associated with infection or bone necrosis due to vertebral compression fracture[1]. IP is usually asymptomatic and is often discovered incidentally on X-rays or CT scans[2,3]. Previous studies found IPs in 10.3% of the para-sacroiliac joints of patients who underwent pelvic CT[4] and in 9.0% of the cervical spines of patients who underwent cervical CT[5]. Although IP is not rare, there are few reports of IP in the cervical rib, scapula, clavicle, humerus, pubis, and acetabulum[3]. To our knowledge, only two cases of IP in the scapula have been previously reported[1,6]. Herein, we report two cases of IP of the scapula.

Case Presentation

Patient 1

A 41-year-old woman presented to an orthopedic clinic with a right thoracic bruise. A plain X-ray revealed a fracture of the right seventh rib and a bone tumor–like lesion in the right scapula. Six weeks later, the patient visited our department. Her medical history and blood test results were unremarkable, and she reported no pain, tenderness, or limited range of motion in the right shoulder. A plain X-ray showed a lobular osteolytic lesion measuring approximately 25 mm in length with a scle-

Correspondence to Yasuyuki Kitagawa, kitayasu@nms.ac.jp
https://doi.org/10.1272/jnms.JNMS.2026_93-202
Received: October 3, 2024; Accepted: November 27, 2024

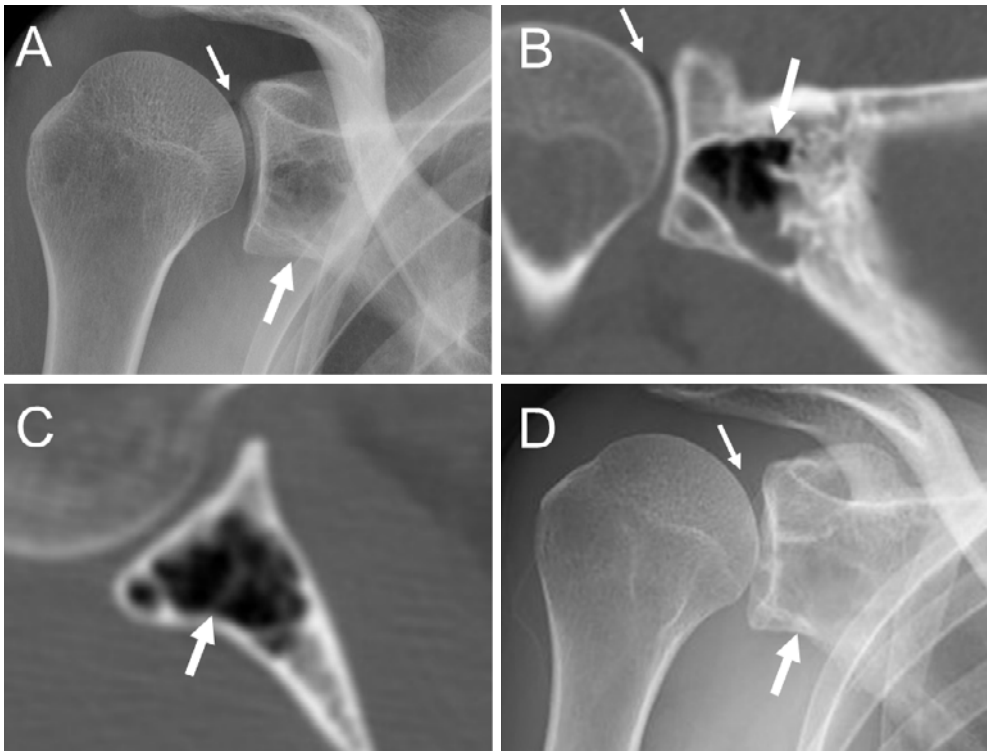

Figure 1 Plain radiographs and CT images of Patient 1
A: A plain X-ray at the initial examination of a 41-year-old woman shows a lytic lesion with a sclerotic rim (thick arrow) in the glenoid to the neck of the right scapula and intraarticular gas (thin arrow). B and C: CT scans at the initial examination show a mixture of gas (minimum CT value, –790 Hounsfield Units [HU]) and soft tissue in the lesion (thick arrow), and intrtaarticular gas (thin arrow). D: A plain X-ray obtained 10 years later shows thickening of the sclerotic rim and decreased internal radiolucency (thick arrow). A small amount of air is present in the shoulder joint (thin arrow).

rotic rim extending from the right scapular neck to the subchondral bone of the glenoid (**Figure 1**). Gas was present in the shoulder joint but not on the unaffected side. CT scanning revealed a mixture of gas (minimum CT value, –790 Hounsfield Units [HU]) and soft tissue in the lesion (**Figure 1**). The lesion was diagnosed as IP. Three weeks later, MRI showed a reduction in the gas-filled areas, low signal intensity on T1- and T2-weighted images, and no contrast enhancement (**Figure 2**). Other regions of the lesion were hypointense on T1-weighted imaging and homogeneously hyperintense on T2-weighted imaging. After contrast administration, the cyst wall showed mild enhancement, suggesting the presence of fluid. The patient remained pain-free throughout the 10-year follow-up period. Plain radiographs obtained 10 years later showed no significant change in size; however, the marginal sclerosis had thickened, internal radiolucency had decreased, and there was a small amount of air in the shoulder joint (**Figure 1**).

Patient 2

A 51-year-old man was diagnosed as having a scapular tumor during a medical checkup and was referred to our department. His medical history and blood test results were unremarkable, and he reported no pain, tenderness, or limited range of motion in the right shoulder. A plain X-ray showed a heterogeneously osteolytic lesion measuring approximately 27 mm in length with a thin rim extending from the right scapular neck to the subchondral bone of the glenoid (**Figure 3**). The X-ray also showed narrowing of the joint space and formation of small osteophytes in the inferior part of the joint; however, there was no gas in the joint. CT revealed the presence of gas within the lesion (minimum CT value, –580 HU), indicating a diagnosis of IP (**Figure 3**). CT also showed a small amount of residual trabecular bone in the lesion, thinning of the subchondral bone adjacent to the lesion, narrowing of the joint space, and intraarticular gas. Four weeks after the initial visit, MRI revealed that the gas-filled area, which appeared as a hypointense signal on T1- and T2-weighted images with no contrast enhance-

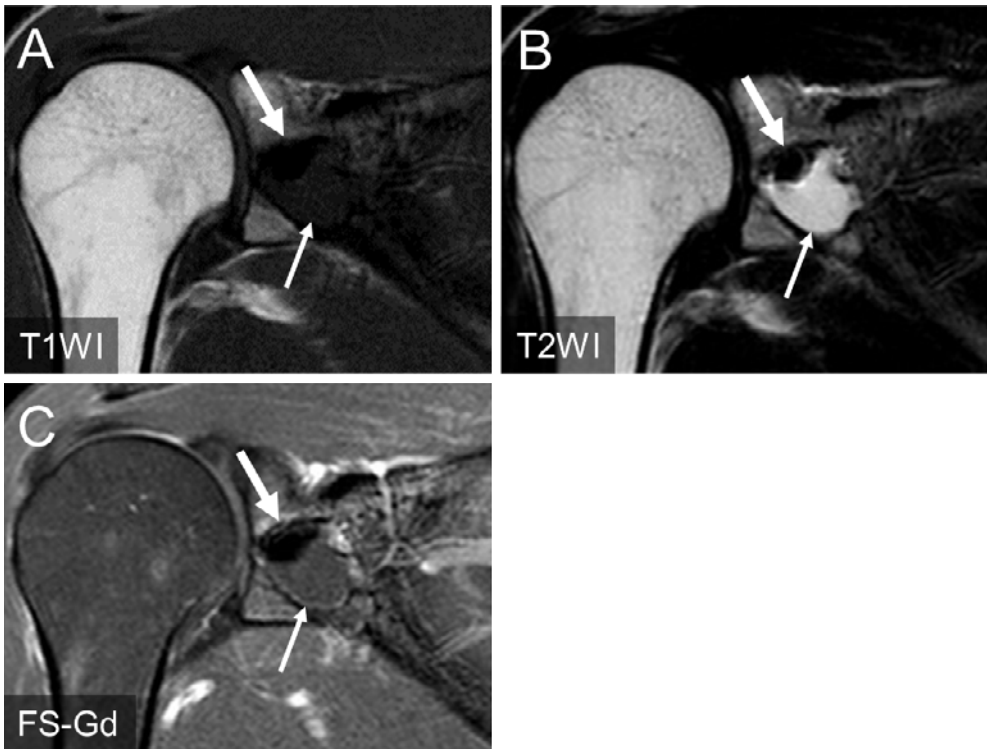

Figure 2 MR images of Patient 1
A, B and C: MRI scans at 3 weeks after the initial visit show a decrease in the gas area (thick arrow). Other areas in the lesion are hypointense on T1-weighted imaging and homogeneously hyperintense on T2-weighted imaging. The cyst wall is weakly enhanced after contrast administration, suggesting the presence of fluid (thin arrow).

ment, had decreased in size (**Figure 4**). The non-gas areas were hypointense on T1-weighted imaging and hyperintense, with scattered dot-like hypointense areas, on T2-weighted images. After contrast administration, only some areas showed enhancement, suggesting the presence of granulation tissue and edematous tissue. At a follow-up visit 6 months later, the patient was pain-free, and a plain X-ray showed reduced radiolucency in the osteolytic area (**Figure 3**).

Discussion

IP prevalence in men is 2.5 times that in women. Mean lesion size is 11.1 mm and mean patient age is 50.4 years[3]. The most common site is near the sacroiliac joint, followed by the spine; however, there are reports of rare cases of IP in the cervical rib, scapula, clavicle, humerus, pubis, and acetabulum[3]. IP is usually asymptomatic and is often discovered incidentally on X-rays or CT scans[2,3]. CT is useful for diagnosis, and characteristic findings include gas attenuation and a thin sclerotic rim[1]. The lesion may contain gas only or a combination of gas, liquid, and soft tissue[7]. Communication between the joint and IP was observed in 20.8% of cases[3]. The differential diagnosis includes infectious osteomyelitis, osteonecrosis, solitary bone cyst, subchondral cyst, intervertebral disc degeneration, and penetrating trauma such as that caused by surgery or arthroscopy[3]. Generally, no treatment is required, but in rare cases surgery may be indicated if the lesion is large enough to pose a risk of fracture[2]. The natural history of IP varies and remains unclear[2]. It has been reported that gas in the cyst may be replaced over time by fluid and eventually by granulation tissue[7].

There are several theories regarding the pathogenesis of IP. One theory suggests that subchondral bone cysts or intraosseous ganglions act as precursor lesions, and when the vacuum phenomenon (VP) occurs in the adjacent joint, gas forms in these precursor lesions. Another theory posits that gas forms as a result of the natural regression of subchondral bone cysts or intraosseous ganglions. A third theory suggests that gas may spread into the bone from a degenerated intervertebral disc that has developed the VP[1,8,9]. After considering factors such as the thick cyst wall, the presence of adjacent intra-shoulder joint gas, the history of rib fracture, and the absence of osteoarthritis, we hypothesize that gas generation in our Patient 1 was due to an intraosseous ganglion rather

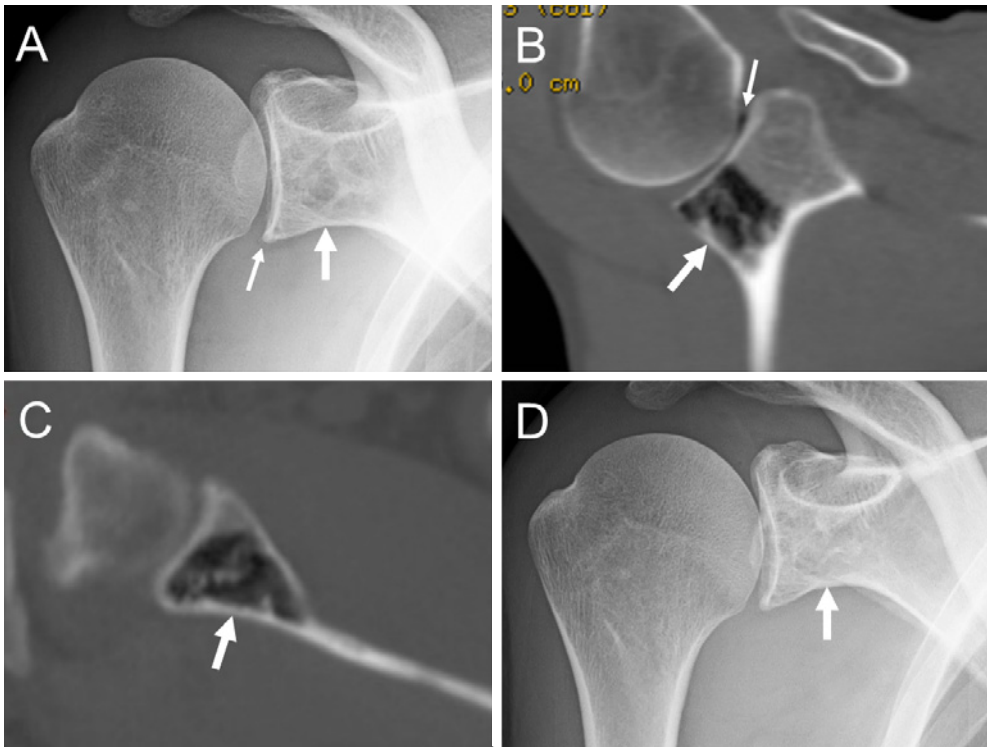

Figure 3 Plain radiographs and CT images of Patient 2
A: A plain X-ray at the initial examination of a 51-year-old woman shows lytic lesion with a thin sclerotic rim (thick arrow) in the glenoid to neck of the right scapula. Narrowing of the joint space and formation of small osteophytes are visible in the inferior area of the joint (thin arrow), but no gas is present in the joint. B and C: CT scans reveal gas in the lesion (minimum CT value, −580 HU) (thick arrow), thinning of the subchondral bone, narrowing of the joint space, and intraarticular gas (thin arrow). D: A plain X-ray obtained 6 months later shows reduced radiolucency in the osteolytic area (thick arrow).

than a subchondral bone cyst. We believe that sudden shoulder joint movement created negative pressure in the joint, leading to gas formation in the intraosseous ganglion. Although imaging did not reveal clear continuity between the joint cavity and bone lesion, the proximity of the lesion to the cartilage suggests that minute continuity cannot be entirely ruled out. Interestingly, X-rays obtained 10 years after follow-up still showed a small amount of intraarticular air.

In contrast, Patient 2 may have had a subchondral cyst as a preceding lesion, as arthropathy was present. A previous study reported that intraarticular gas visible on CT during shoulder elevation is present in approximately 10% of healthy individuals[10]. Although this indicates that gas accumulation itself has little pathological significance, it suggests that negative pressure sufficient to cause VP can easily occur in the joint. In this patient, we speculate that VP developed in the joint, leading to gas formation in the subchondral cyst.

VP occurs in joints when a sudden force on the joint degrades the articular surfaces, resulting in negative pressure and the release of gases, mainly nitrogen, from soft tissues. Although most cases are caused by negative pressure in the joint cavity induced by trauma or osteoarthritis, VP can also be caused by normal joint movement or traction[11]. The shoulder joint is a ball-and-socket joint, and the articular surface accounts for only 25% of the humeral head, making it the most unstable joint with the greatest range of motion in the human body[12]. Even in healthy people, intraarticular VP was observed in approximately 10% and 50% of shoulder joints in the elevated and externally rotated positions, respectively, but not in the neutral position[10,13].

To our knowledge, only two cases of IP in the scapula have been previously reported[1,6]. The first was a 71-year-old man with IP that was discovered incidentally on CT[1]. The lesions were located in the left scapular glenoid and were identified as air-containing ovoid lesions measuring 13 × 6 mm and 9 × 5 mm with thin sclerotic rims. Because osteoarthritis was mild, the authors speculated that the preceding lesion was more likely to have been an intraosseous ganglion than a subchondral bone cyst. They

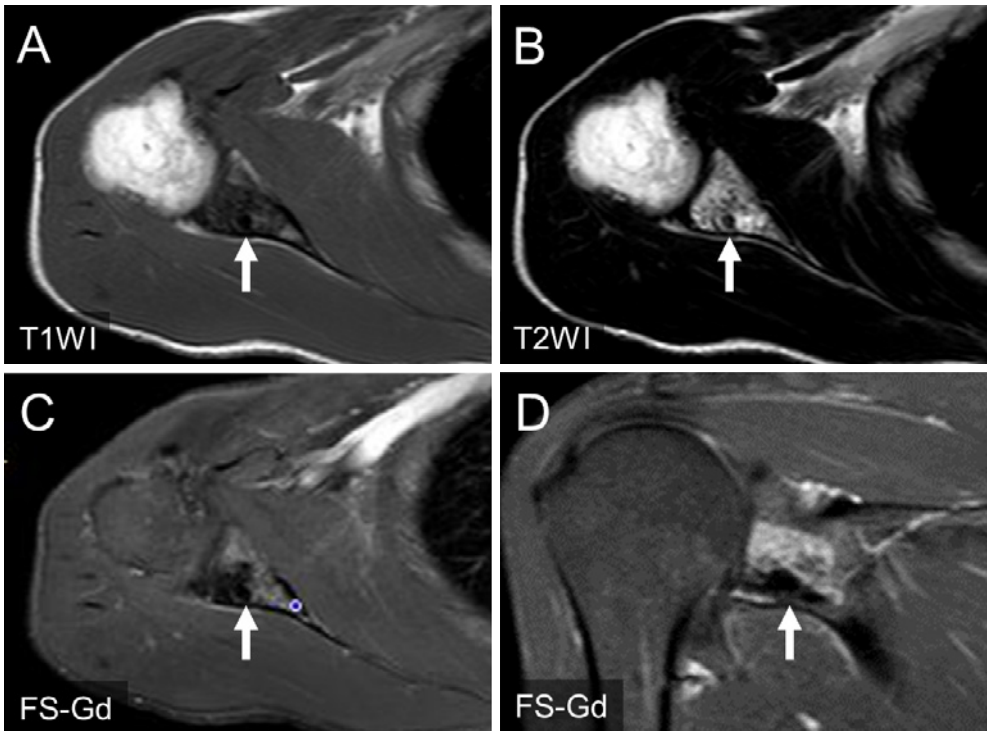

Figure 4 MR images of Patient 2
A, B, C and D: MRI scans at 4 weeks after the initial visit show shrinkage of the gas-filled area (thick arrow). The non-gas areas exhibited low signal intensity on T1-weighted imaging and high signal intensity with scattered dot-like low signal areas on T2-weighted imaging. After contrast agent administration, only some areas are enhanced, suggesting the presence of granulation tissue and edematous tissue.

also hypothesized that the presence of bone ganglion and degenerative changes in the shoulder joint was involved in the development of IP. The other case was a 40-year-old man with a lesion in the glenoid of the scapula, with a suspected preceding intraosseous ganglion[6].

A previous study of the relationship between IP and intraarticular VP reported that four of five cases of vertebral IP were associated with VP of the intervertebral disc and that all five cases of IP near the facet joint were associated with VP of the facet joint[5]. Another study reported that six of eight cases of sacral IP were associated with intraarticular VP and that two had communication with intraarticular VP. All eight of these patients had osteoarthritis[14].

In conclusion, we reported two rare cases of IP of the scapula. To our knowledge, these are the third and fourth reported cases involving the scapula. Both lesions were located in the scapular neck to the glenoid, adjacent to the shoulder joint, and had a sclerotic rim. CT was useful for diagnosis in both cases. We speculate that gas was generated when articular VP developed in a preceding intraosseous ganglion or subchondral cyst.

Author Contributions: The authors participated in the clinical management of the patient, data acquisition, analysis, and manuscript drafting. All authors approved the final version of the manuscript and agreed to be accountable for all aspects of the work.

Funding: None.

Conflict of Interest: None declared.

Declaration of Generative AI and AI-Assisted Technologies in the Writing Process: None.

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