Abstract
Eosinophilic granuloma (EG) is a benign tumor-like condition which is characterized by a clonal proliferation of Langerhans-type histiocytes in the bone or lung [1, 2]. The causes and pathogenesis of EG are unknown. In spite of extensive genetic studies or virologic analyses, apparent genetic error or infectious agents have not been found [3, 4]. The radiographic features of EG is quite different depending on the phase of the disease and the site of involvement [3, 5]. A status of EG in the

Introduction
Eosinophilic granuloma (EG) is a benign tumor-like condition which is characterized by a clonal proliferation of Langerhans-type histiocytes in the bone or lung [1, 2]. The causes and pathogenesis of EG are unknown. In spite of extensive genetic studies or virologic analyses, apparent genetic error or infectious agents have not been found [3, 4]. The radiographic features of EG is quite different depending on the phase of the disease and the site of involvement [3, 5]. A status of EG in the
bone is divided into acute and chronic phases [5, 6, 7]. The acute phase of EG is a destructive, osteolytic lesion and has poor margins. Therefore it is difficult to differentiate from a malignant tumor such as Ewing’s sarcoma, or acute osteomyelitis [5]. On the other hand, the chronic phase of EG is a well-defined lesion, and the radiological features are similar to those of chronic osteomyelitis (Brodie’s abscess) or a benign bone tumor [5, 8].

EG is a local condition of Langerhans cell histiocytosis (LCH) and make up 80% of LCH in children [5]. The systemic condition of LCH is a multisystem life-threatening disorder. For example, Hand-Schüller-Christian disease is defined as a triad of exophthalmos, diabetes insipidus and osteolytic lesions of the skull, and Letterer-Siwe disease shows hepatosplenomegaly, lymphadenopathy, skin rash, fever, anemia and thrombocytopenia [1, 4]. Though these three diseases are quite different in disease expression pattern and their prognosis, the histology is similar to each other and indistinguishable [4]. The incidence of EG is estimated at 0.05–0.5 per 100,000 and 90% of EG occurs under the age of 15 years old [3, 4, 5]. Male has twice as high incidence rate as female [8]. Flat bones are involved in 70% of cases and long bones in 30% [8]. Among the flat bones, the skull is most frequently involved, and the pelvis, vertebrae, mandible, and ribs are followed in decreasing order [1, 5]. We report 3 children’s cases of EG in the pelvis which showed quite different radiological features and clinical courses.

Case reports

Case 1
A 6-year-old boy was referred to our hospital with pain in the right thigh and limping for 2 months. Physical examination indicated tenderness over the right hip joint without swelling. The motion of the joint was slightly restricted due to pain. Plain radiographs of the pelvis showed a slightly defined osteolytic lesion in the right acetabulum (Figure 1A). Blood test demonstrated normal white blood cell (WBC) count (7,800 /μl), eosinophils (4.7%), and slight increased erythrocyte sedimentation rate (ESR) (21 mm/h). Computed tomography (CT) showed a 3.0 × 2.0 cm osteolytic low density lesion in the right acetabulum with a slightly defined margin (Figure 1B). Magnetic resonance imaging (MRI) revealed that the mass had lower signal intensities on T1-weighted images and higher signal intensities on T2-weighted images than the bone marrow (Figure 1C, D). The right ilium also had lower signal intensities on T1-weighted images (Figure 1C). The mass extended into the lateral soft tissue of the pelvic bone on T2-weighted images (Figure 1D). Based on these examinations, an initial radiological diagnosis was acetabular osteomyelitis and an open biopsy was performed through the anterior approach. Histological examination revealed mixed cellular infiltrate with histiocytes, eosinophils, lymphocytes, and macrophages, which led to the diagnosis of EG (Figure 1E). Because pain was relieved in a week after the biopsy, no further treatment was performed. The plain radiograph showed the signs of healing in 3 months after the biopsy (Figure 1F), and complete healing was observed in a year.
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Case 2

A 4-year-old boy was referred with a month history of pain and limping in the left lower limb. Physical examination revealed tenderness over the left hip joint without swelling. The motion of the joint was not restricted. Blood test showed normal WBC count (8,520 /μl), eosinophils (5%), and slightly increased ESR (35 mm/h). Plain radiographs showed a 3.0 × 2.5 cm osteolytic lesion with slight marginal sclerosis in the left acetabulum (Figure 2A). A complete bone scan with 99mTc Technetium showed an increased uptake in the left acetabulum (Figure 2B). The lesion and the surrounding area in the pelvic bone had lower signal intensities on T1-weighted images (Figure 2C). The lesion and the lateral soft tissue of the pelvic bone (arrow) demonstrated higher signal intensities. E: Histological examination indicating a mixed cellular infiltrate with histiocytes, eosinophils, lymphocytes, and macrophages. Arrows show eosinophils. F: Three months after the open biopsy. Radiographic signs of healing appeared.

Case 3

A 2-year-old boy was referred with a limping in the left leg for a month. On physical examination, motion of the joint was not restricted and neither swelling nor tenderness was observed. WBC and ESR increased (11,460 /μl and 30 mm/h, respectively). Eosinophils were within the normal range (3%). Plain radiograph
of the pelvis revealed an osteolytic lesion with ill-defined margins in the ilium (Figure 3A). CT showed an osteolytic lesion in the ilium extending into the medial retroperitoneal space (Figure 3B). The left ilium showed lower signal intensities on T1-weighted images (Figure 3C) and higher signal intensities on T2-weighted images which extended into the medial retroperitoneal space and the lateral gluteal muscles (Figure 3D). Bone scan with $^{99m}$Tc-Technetium showed a massive uptake in the left pelvis (Figure 3E). An initial radiological diagnosis was Ewing’s sarcoma and CT-guided needle biopsy was performed through the posterior aspect of the left ilium. The histological examination demonstrated a cellular lesion consisting of numerous histiocytes and eosinophils. The lesion was well-defined after the biopsy, but gradually enlarged in a month (Figure 3F). CT also confirmed enlargement of the lesion, but the adjacent sacrum was not involved (Figure 3G). MRI demonstrated that the mass and soft tissue abnormalities became enlarged (Figure 3H, I). Curettage was carried out immediately. His pain and limping subsided gradually after the operation. However, the plain radiograph taken after 1 year later showed that the osteolytic lesion of the left pelvis remained (Figure 3J).

Discussion
The radiographic feature of EG is completely different depending on the phase of the disease and the site of involvement [3, 5]. A status of EG in the bone is divided
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into acute and chronic phases [5, 6, 7]. The acute phase of EG shows osteolysis with poorly defined margins and the chronic phase of EG shows well-defined margins. Case 3 is consistent with the acute phase and case 1 and 2 are the chronic ones. In the pelvis, the lesion may present as poorly defined areas of osteolysis in the acute phase that become progressively circumscribed as they mature in the chronic phase [4].
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MRI is a sensitive, but nonspecific modality to detect bone marrow involvement and a soft tissue mass [4, 5, 9]. T1-weighted images are useful for demonstrating bone marrow involvement and T2-weighted images for indicating a soft tissue mass [4, 5, 9]. These changes are considered as edema of bone marrow and soft tissues [4]. Edema of adjacent bone marrow and soft tissue is particularly seen in the acute phase lesions [5]. Chronic phase lesions show decreased signal intensity on T2-weighted images, indicating re-ossification of the osteolytic lesions with resolution of the soft tissue mass [4]. The acute phase is defined as the tumor invading the surrounding soft tissue but the chronic phase is localized as itself [8]. Case 3 showed an extensive, irregular soft tissue mass in the medial retroperitoneal space and the lateral gluteal muscles. On the other hand, the mass in the case 1 and 2 was localized.

Vertebral lesions show a symmetrical flattening of the vertebra with intervertebral disc space preservation, which are called as “vertebra plana”, and easily diagnosed only by plain radiographs [4]. However, it is very difficult to diagnose from radiological examinations in the pelvis. Plain radiograph or MRI is a very sensitive modality to detect the lesions, but it is not specific because of the radiological diversity of EG. We could not distinguish EG from malignant bone tumors such as Ewing’s sarcoma or acute osteomyelitis in the case 3 and a chronic osteomyelitis or benign bone tumors in the case 1 and 2. Therefore, not only plain radiograph and MRI but also open or needle biopsy is indispensable to arrive at the correct diagnosis of EG.

In spite of good prognosis of EG, various therapeutic approaches have been proposed. The effectiveness of curettage and bone grafting, local injection of corticosteroids, irradiation, and chemotherapy has been reported [1, 10–13]. Because the incidence of EG is low (1% of total primary bone tumor) and the trend to spontaneous healing is high, it is very difficult to assess the true efficacy of these therapies. Treatment of EG affecting the pelvis is usually accomplished by curettage of the affected site and bone grafting [4]. On the other hand, spontaneous healing of EG after open or needle biopsy has been reported by several authors [2, 4, 5]. Case 1 and 2 showed a pain relief in a few weeks after biopsy and spontaneous healing in a few months. In this respect, a careful observation after open or needle biopsy is thought to be one of the treatment options for EG. Some of EG may resolve spontaneously, and the other will persist or expand after biopsy [12]. Case 3 showed an osteolysis with ill-defined margins and large soft tissue mass which extended into medial retroperitoneal space and the lateral gluteal muscles. This case did not resolve spontaneously. Howard et al. reported a similar case of EG in the pelvis as the case 3 which showed prominent osteolysis with ill-defined margins and large soft tissue edema extending into both side of the bone on T2-weighted images. This case did not show any tendency to resolve after biopsy, and additional curettage was carried out [14]. EG with acute radiological features such as osteolysis with ill-defined margins and large soft tissue edema on T2-weighted images may progress after open or needle biopsy. The chronic form of EG with well-defined margins and localized soft tissue mass on T2-weighted images may resolve spontaneously. We need further attention for EG especially with acute radiological features.
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Corresponding author:
Masahito Hatori
E-mail: mhato@mail.tains.tohoku.ac.jp
Phone: +81-22-7177245
Fax: +81-22-7177248
