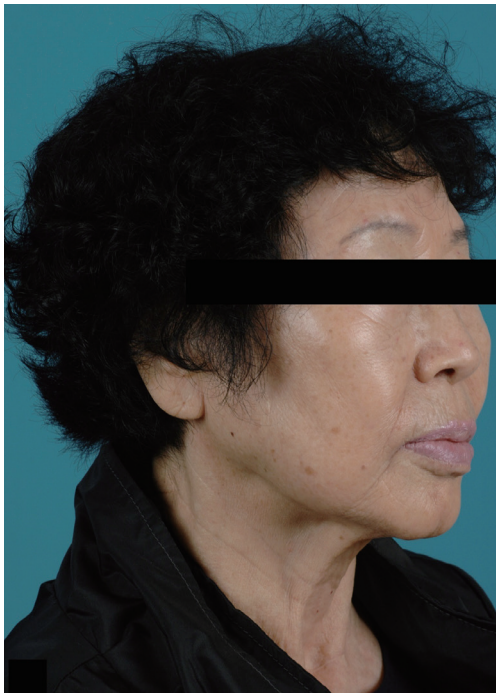

Fig. 4.
Postoperative 3-month image. There is no evidence of recurrence.

approaches have been used, including a preauricular approach with or without extension to a temporal, hemicoronal incision, submandibular approach, preauricular and intraoral approach, and modified Blair incision. The preauricular approach, either on its own or in combination with cervical incisions, is the most popular approach [4]. However, the conventional surgical approach seems to be limited by technical difficulties in achieving adequate exposure. In terms of resection, the conventional approach has been condylectomy in cases of condylar osteochondroma, particularly in older case reports [3-5]. More recently, a more conservative approach to resection has been frequently reported. The recurrence rate of osteochondroma generally is 2% [1,4]. It is important to remove the tumor completely under direct vision. There is no reported endoscope-assisted complete excision of osteochondroma at the mandibular angle yet. In this rare case of osteochondroma of the mandibular angle in an 81-year-old female, the painless mass was on her mandibular angle for 20 years. In this case, with an endoscope-assisted oral incision, we were able to achieve good exposure, followed by a successful outcome. There was no evidence of recurrence at 6 months after surgery.

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Langerhans Cell Histiocytosis with Frontal Bone Indentation by an Adjoining Primary Soft Tissue Lesion in a 17-Month-Old Asian Male Child

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Langerhans cell histiocytosis (LCH) is a rare disease that represents a clonal proliferation of pathologic Langerhans cells [1]. Although its clinical manifestations range from isolated bone lesions to multisystem disease, the pathological finding is

uniform irrespective of the disease manifestation. Besides their characteristic morphology, Langerhans cells are further identified histochemically by the presence of the CD1a and S-100 surface markers and electro-microscopically by the presence of Birbeck granules [2]. Pathological findings are obligatory for diagnosis. LCH usually occurs in young children with a peak from 1 to 4 years [3]. Soft tissue involvement is usually the secondary extension of an adjoining primary bone lesion, while the secondary involvement of an adjacent bone by a primary soft tissue LCH lesion is rare [4]. Here, we report the case of a 17-month-old Asian male child with a solitary primary soft tissue LCH lesion in the right forehead area with secondary bone destruction by indentation. This 17-month-old male child was referred to our institution for a forehead mass that rapidly enlarged in three weeks. This child initially presented at an outside clinic, and an ultrasonographic (US) evaluation was performed. The US revealed a soft tissue mass with a diameter of 22 mm and with bone erosion and destruction on the right forehead. LCH was suspected, and the patient was referred to our institution. Initial recognition of the mass was at three weeks before admission. Physical examination revealed a 3.0×3.0 cm tender and fixed mass over the right forehead. Computed tomography (CT), fluorodeoxyglucose positron emission tomography (^{18}F-FDG-PET)/CT, and skull radiography imaging studies were performed before open surgery (Figs. 1, 2). In December 2011, the mass was extirpated under general anesthesia. On the operative field, we observed an irregularly shaped firm mass measuring 2×3 cm, located in the subcutaneous layer with periosteal inflammation and thinning of the frontal bone without dural exposure. To reconstruct the thinning of the frontal bone, we performed an allograft with a demineralized bone matrix and an osteoconductive bone void filler. Pathologic findings showed that under high magnification (×400), the tumor cells were mostly large monocytes with abundant cytoplasm and occasionally central grooved nuclei, admixed with numerous eosinophils. The presence of Langerhans cells was confirmed by CD1a and S-100 immunohistochemistry (Figs. 3, 4). To identify whether the tumor origin was the soft tissue and the bone, we conducted a pathologic study of the adjoining periosteum. The histologic study revealed mild chronic inflammation with fibrosis; this result confirms the diagnosis of primary soft tissue LCH. After surgery, this child was followed-up at Samsung Medical Center every 6 months with a bone scan (Fig. 5), and the 1-year follow-ups were conducted at our institution. During the follow-up period of 3 years, no evidence of recurrence was found. LCH, previously known as histiocytosis X, is a histiocytic disorder of unknown etiology that is characterized by the clonal proliferation of dendritic cells in various organs and tissues. LCH has a variable clinical presentation and course, ranging from a solitary, often

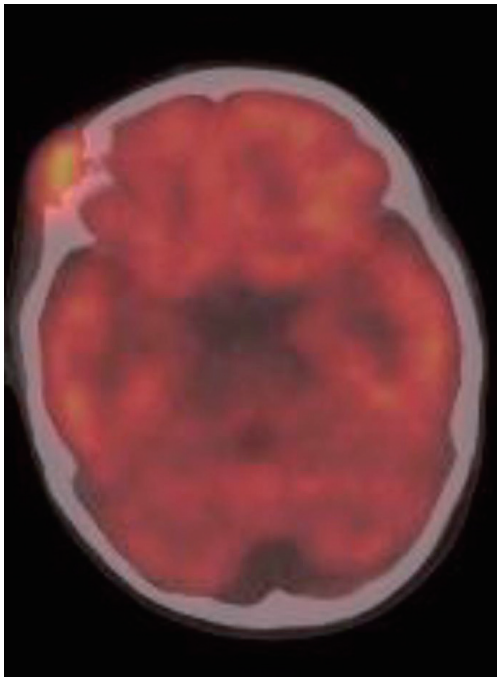

Fig. 1.
Fluorodeoxyglucose positron emission tomography (^{18}F-FDG PET) imaging study. Demonstrating a 5×1.4 cm focal FDG uptake lesion with right frontal bone destruction in the right forehead area.

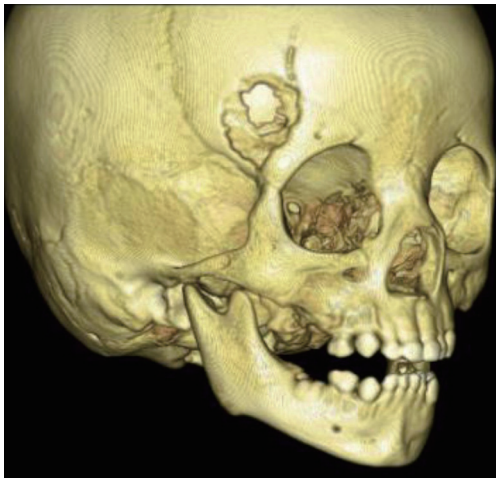

Fig. 2.
Computed tomography imaging study. Demonstrating the irregularly beveled marginated lytic bone defect involving the right frontal skull vault including the inner table and the superolateral orbital walls and just above the frontozygomatic suture.

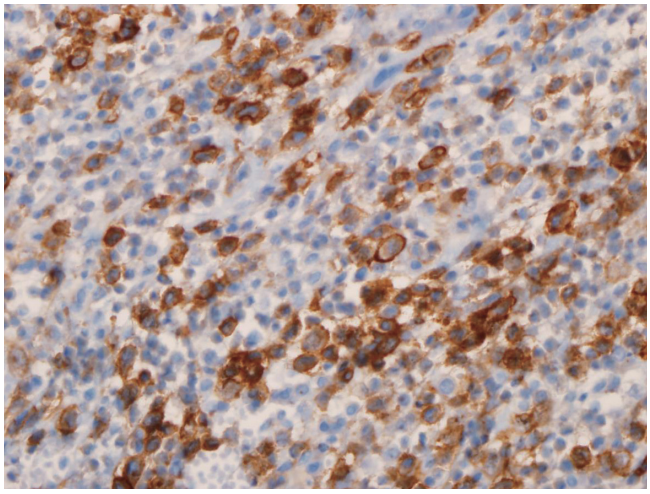

Fig. 3.
The immunohistochemical stains for CD1a. Strong positive in the cytoplasm of tumor cells (×40).

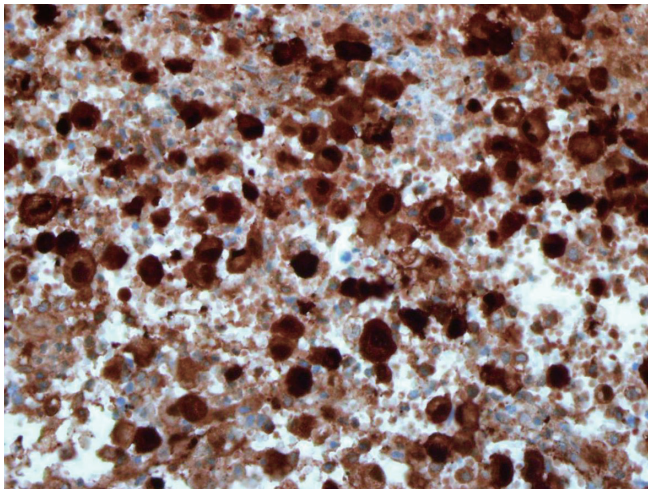

Fig. 4.
The immunohistochemical stains for S-100 protein. Positive cells with elongated nuclei (×40).

spontaneously resolving, bone lesion to a fatal multicentric and multiorgan system disease. LCH lesions can involve any organ in the body; however, bones, skin, and lungs are the most common sites of involvement. Soft tissue involvement is usually due to the extension of an adjoining primary bone lesion [4]. In our case, LCH with a soft tissue origin affected the adjoining bone by indentation. Before surgical exploration, the origin of the mass was difficult to identify on the basis of radiologic findings. To identify whether the tumor origin was the soft tissue or the bone, we conducted a pathologic study of the adjoining periosteum. The histologic study revealed mild chronic inflammation with fibrosis; this result confirmed the diagnosis of primary soft tissue LCH. There are multiple treatment modalities for LCH: solitary, easily accessible lesions can be surgically extirpated, and intralesional corticosteroid application and radiotherapy are also possible options [5]. However, clinical guidelines for the treatment of LCH are still under development. In our patient, timely surgical extirpation of the mass was performed and no evidence of recurrence was found during the 3-year follow-up period. Here, we presented the case of a 17-month-old child with a solitary primary soft tissue LCH lesion, which influenced the adjacent bone by indentation. Due to the rarity of LCH that involves both the soft tissue and the bone, it is difficult to identify the tumor origin. In this case, LCH with an ambiguous origin was completely resolved by surgical extirpation, and the primary soft tissue origin of the tumor was confirmed by a histologic study.

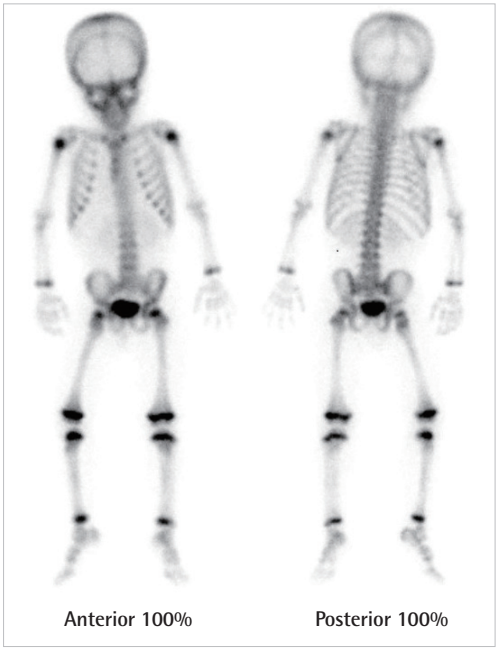

Fig. 5.
Follow-up bone scan taken 2 years after surgery. No evidence of recurrence and bony lesion.

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Lymphoepithelioma-like Carcinoma of the Skin in the Cheek with a Malignant Metastatic Cervical Lymph Node

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Since first described by Swanson et al. in 1988, lymphoepithelioma-like carcinoma of the skin (LELCS) has been considered a rare malignant skin tumor. It is usually caused by sun exposure and is therefore most prevalent on the skin of the head and neck in elderly individuals. Although the histologic etiology of LELCS is still unclear, it is known to have a slight tendency toward local recurrence and extremely low metastatic potential. Approximately 70 cases have been reported in the literature to date [1]. Among them, only a few reports have described local recurrence or metastasis to lymph nodes, and only one reported death due to distant metastasis of LELCS. In Korea, two LELCS cases have been reported in the Korean language. However, both cases had no local recurrence or lymph node metastasis nor any long-term follow-up data. We report the first case of LELCS with a malignant metastatic cervical lymph node in a Korean. A 71-year-old male with a skin lesion of the right cheek, which appeared several months prior to presentation, visited our hospital. Clinically, the lesion presented as soft, mobile over the underlying tissues, and painless, with a diameter of 5 mm and an erythematous macule with central eschar caused by a punch biopsy at an another hospital (Fig. 1). He did not have any palpable lymphadenopathy or nasopharyngeal symptoms. The slide from the other hospital was re-evaluated in our hospital; poorly differentiated carcinoma was identified in the dermis. Magnetic resonance imaging (MRI) showed a focal defect lesion approximately 5 mm in size surrounding the enhancement with an ambiguous impression suggesting either tumor enhancement or a post-biopsy reaction (Fig. 2), and a whole-body positron emission tomography (PET) scan showed skin thickening with focal hypermetabolism in the right cheek. In ultrasonography (USG), palpable and abnormal lymph nodes were not observed. Surgical treatment was performed with wide excision (surgical

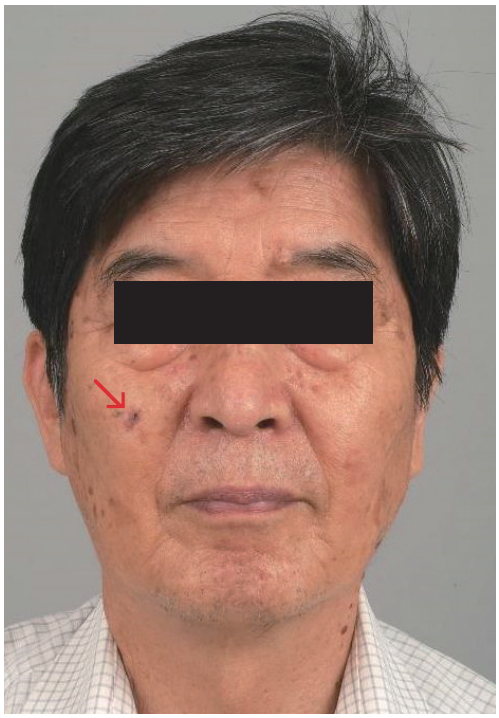

Fig. 1.
Preoperative photograph of the patient. An erythematous macule with central eschar (red arrow) can be observed in right cheek.

Images