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Treatment of Osteofibrous Dysplasia and Associated Lesions

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Purpose: To report long term treatment outcomes of osteofibrous dysplasia and association with adamantinoma. **Patients and Methods:** From January 1984 to July 2001, 14 patients with osteofibrous dysplasia were followed for an average of 108 months (78 to 260 months). Our patient group consisted of 6 men and 8 women, with a mean age of 13.9 years (2 to 65 years). We reviewed the clinical and pathological features of all 14 patients. **Results:** Thirteen patients had a lesion in the tibia, while one patient had lesions in both the tibia and the fibula. Initial treatments were observation after biopsy (6 patients), curettage with or without a bone graft (3 patients), resection followed by a free vascularized fibular bone graft (4 patients), or resection and regeneration with the Ilizarov external fixation (1 patient). Curettage was performed on 6 patients due to recurrence or progression after the initial treatment. Among these patients, one was diagnosed with AD from the biopsy of the recurrent lesion. This patient was further treated by segmental resection and pasteurization. After the initial pathology slides of the 13 patients were reviewed with immunohistochemical cytokeratin staining, one patient diagnosis was changed from osteofibrous dysplasia to osteofibrous dysplasia-like adamantinoma. **Conclusion:** Some patients with osteofibrous dysplasia require close observation because of the high association risk between osteofibrous dysplasia and adamantinoma, Immunohistochemical staining may be helpful in differentiating these two diagnoses.

Key Words: Osteofibrous dysplasia, osteofibrous dysplasia like adamantinoma, adamantinoma

INTRODUCTION

Osteofibrous dysplasia (OFD) is a rare developmental condition of childhood, which almost exclusively affects the tibia. The tumor affects children in their first decade and ceases progression with the termination of growth. The important differential diagnosis of OFD should include monostotic fibrous dysplasia and adamantinoma (AD). AD is a low-grade malignant epithelial neoplasm of the cortical bone of the tibia, which shares similar radiological features to OFD and may be difficult to differentiate histopathologically. Some recent reports have suggested that OFD is a precursor of a neoplastic process leading to AD,[1-4] and others have suggested that OFD is a residue of spontaneously regressing AD.[5-7]

While wide excision or amputation is required for the treatment of AD, observation until growth is completed is recommended for OFD.[8-11] Recent reports recognized OFD-like AD has a small nest or strand of epitheloid-originated cells, which makes the proper disease management more difficult.[2,4,5,12-16]

The purpose of our study is to review the long-term clinical results of patients diagnosed and treated for OFD and to present the association between three disease entities.

MATERIALS AND METHODS

Patients data and clinical features

Fourteen patients diagnosed with OFD from January 1984 to July 2001 were reviewed retrospectively. The patient group consisted of 6 men and 8 women, with a mean age of 13.9 years (2 to 65 years). Thirteen lesions involved the tibial shaft and 1 lesion involved both the tibia and the fibula simultaneously. Twelve patients presented

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with a palpable mass, 8 patients complained of pain and tenderness, and 7 had anterior bowing of the tibia. One patient was found without any symptoms. An incisional biopsy was performed on all patients. The patients were followed for 78 to 260 months (average, 108 months), postoperatively. Follow-up radiological findings, such as eccentric, well-marginated, and multi-located, expansive radiolucent lesions with destructive cortexes were checked for recurrences. Diagnosis was made by pathological confirmation, regardless of the radiological findings. After biopsy, the radiological follow-up was established every 3 months for the first year, and then every 6 months if no disease progression was found. Operative treatment was performed in the following cases: if the disease progressed within a three months interval, if the lesion was so large that impending fracture was a risk, or if the recurrent pathological fracture was present. Wide resections and reconstructions were carried out if the lesion involved the entire tibia or if the tibia showed severe anterior bowing. In other cases, subperiosteal curettage or en bloc excision was performed. To prevent pathological fractures, the patients were advised to restrict activity and patients wore braces if needed. Follow-up radiographs were taken regularly to check for recurrences. The recurring lesions were biopsied. Slow-growing lesions were managed with observation, and large lesions in patients with completed growth were treated with curettage.

Histological examination

Fourteen cases were retrieved from the routine surgical pathology procedure. The specimens from each tumor were fixed in 10% formalin and embedded in paraffin. Routine HE and immunohistochemical cytokeratin (CK) staining was performed on the fixed sections.

Immunohistochemistry

Three-micron-thick sections were placed on saline-coated slides, deparaffinized, immersed in phosphate-buffered saline (PBS) containing 0.3% (v/v) hydrogen peroxide, and microwaved in 10 mmol/L sodium citrate buffer, pH 6.5, for 15 min at 700 W. After blocking with 1% (w/v) bovine serum albumin in PBS containing 0.05% (v/v) Tween-20 for 30 min, the slides were incubated at 4℃ overnight with primary antibody for cytokeratin (DAKO Cytomation, Glostrup, Denmark; 1 : 300). Immunoperoxidase staining was performed using the LSAB universal kit. For negative controls, antibody was replaced by equivalent amounts of subtype-matched normal mouse IgG for the monoclonal antibodies, and rabbit immunoglobulin fraction for the polyclonal antibodies. The final reaction product was visualized after dissolving 6 mg 3,3′-diaminobenzidine tetrachloride in 10 mL 0.05 mol/L Tris buffer (pH 7.6) containing 0.1 mL 3% hydrogen peroxide for 5-20 min.

The positive immunoreactivity of cytokeratin was only valid along the cytoplasmic border.

RESULTS

Among the fourteen OFD patients, 6 patients were managed only by observation and another 8 patients underwent surgery. Among the 8 patients who had surgery, 7 patients underwent surgical resection during the course of the follow-up, but one patient required immediate resection after the initial biopsy because the lesion involved the entire tibia and the tibial shaft had severe anterior bowing. Six patients without surgical intervention remained disease-free throughout the follow-up period. Out of these 6 patients, 5 were over 14 years-old at the time of diagnosis, which is past the active growth period, and 1 patient showed no progression in the 7 years of follow-up, even though he was only 7 years old at the time of diagnosis. Among the 8 patients who had surgery, 2 patients underwent subperiosteal curettage, 1 patient had an en bloc excision. The remaining 5 patients underwent segmental resection followed by reconstruction with free vascularized contralateral fibular bone grafts in 4 patients and reconstruction with osteogenesis by internal transport using the Ilizarov external fixation apparatus. Among the 2 patients who had subperiosteal curettage, one achieved complete remission and the other had slow disease progression, which only required close observation. A 65-year-

old man who had an en block excision of a small lesion at the proximal shaft of his tibia acquired complete remission.

All five patients who had segmental resections had recurrences. One case (case 6 in Table 1) had a segmental resection and reconstruction with the Ilizarov external fixation apparatus and then underwent subperiosteal curettage and bone grafts for the recurrent lesion, a treatment course that ultimately achieved complete disease remission. A 6-year-old boy (case 2 in Table 1) to whom wide resection was not appropriate because his distal tibial lesion was abutting on the growth plate, received a segmental intralesional resection followed by reconstruction with a free vascularized fibular bone graft. This patient experienced recurrence 4 times near the growth plate and was treated with subperiosteal curettage, 3 bone grafts, and a pasteurization. He has been free of recurrence for the following 13 years.

One female patient (case 1 in Table 1) who had received free vascularized fibular bone grafts had a recurrence of the osteolytic lesion at the margin of the grafted fibula and remaining tibia. The resection and reattachment were performed after pasteurization of the recurrent lesion. The pathology of the recurrent lesion showed large nests of epithelial cells that are typical of AD. The initial pathological slide was reviewed and stained for cytokeratin by immunohistochemistry. Scattered cytokeratin positive epithelial cells were found within the loose fibrous tissue of the initial biopsy. Some of these pieces are compatible with OFD-like AD.

After this case, we reviewed the initial and recurred biopsy specimens from all OFD patients. Among the 13 patients available for review, one patient had a diagnosis altered to OFD-like AD (case 6 in Table 1). Two patients who were reassessed as an adamantinoma or an OFD-like AD were free of recurrence after the excision of the recurred lesion.

Table 1. Summary of Cases

Case No.	Age (yrs)	Sex	Site	Initial pathology	Initial treatment	Outcome (Total recurrence)	F/U (yrs)	Further treatment
1	13	F	Tibia	OFD	S/R & FVFG	Recur (1) → classic AD	19.6	Excision & Past
2	6	M	Tibia	OFD	S/R & FVFG	Recur (4)	19.7	Cur & BG ⇒ Past
3	4	M	Tibia	OFD	S/R & FVFG	Recur (1)	2.5	Cur & BG
4	8	F	Tibia	OFD	S/R & FVFG	Recur (1)	17.3	Excision & Past
5	16	F	Tibia	OFD	Bx & Observation	Non-progression	3	
6	2	M	Tibia	OFD	S/R & E/F wit Ilizarov	Recur (1) → OFD like AD	13.5	Cur & BG
7	18	F	Tibia	OFD	Bx & Observation	Regression	7	
8	7	M	Tibia	OFD	Bx & Observation	Non-progression	6.5	
9	12	M	Tibia	OFD	Bx & Observation	Slowly progression	5.5	Observation
10	16	F	Tibia & Fibula	OFD	Bx & Observation	Non-progression	6.3	
11	3	F	Tibia	OFD	Curettage	Slowly Progression	6.5	Observation
12	11	F	Tibia	OFD	Cur & BG	Healing	5.9	
13	14	F	Tibia	OFD	Bx & Observation	Non-progression	6.9	
14	65	M	Tibia	OFD	En bloc excision	Healing	6.5	

S/R, segmental resection; FVFG, free vascularized fibular graft; E/F, external fixation; Bx, biopsy; BG, bone graft; Cur, curettage; Past, pasterization; AD, adamantinoma; F/U, follow up.

Case 1

A girl aged 12 years and 11 months (case 1 in Table 1) presented with severe anterior bowing of her left tibia (Fig. 1A). Under the diagnosis of OFD, a wide resection and reconstruction with a 21 centimeter, free vascularized fibular bone graft was performed (Fig. 1B). The pathological finding showed proliferation of fibrous tissue surrounding osseous trabeculae rimmed by active osteoblasts and zonal architecture. Recurrence occurred 14 years after the operation (Fig. 1C), and the recurrence was treated by resection and pasteurization (Fig. 1D). The pathological finding on the recurred lesion indicated that it was compatible with classical AD with abundant nests of epithelial cells (Fig. 1E). In contrast, the initial pathology was predominantly composed of fibro-osseous stromal tissues without epithelial cells. Using an immunohistochemical analysis, CK stained strongly along the cytoplasmic border of tumor cells (Fig. 1F). In addition, the immunohistochemistry from the initial biopsy revealed some scattered CK-positive epithelial cells within the loose fibrous tissue, a finding which was compatible with an OFD-like AD.

Four years and 11 months after the operation to treat the recurrence (19 years and 7 months after the initial operation), we found no evidence of recurrence or distant metastasis. The patient shows no limitation in ambulation, with full range of motion in the knee and ankle joints (Fig. 1G).

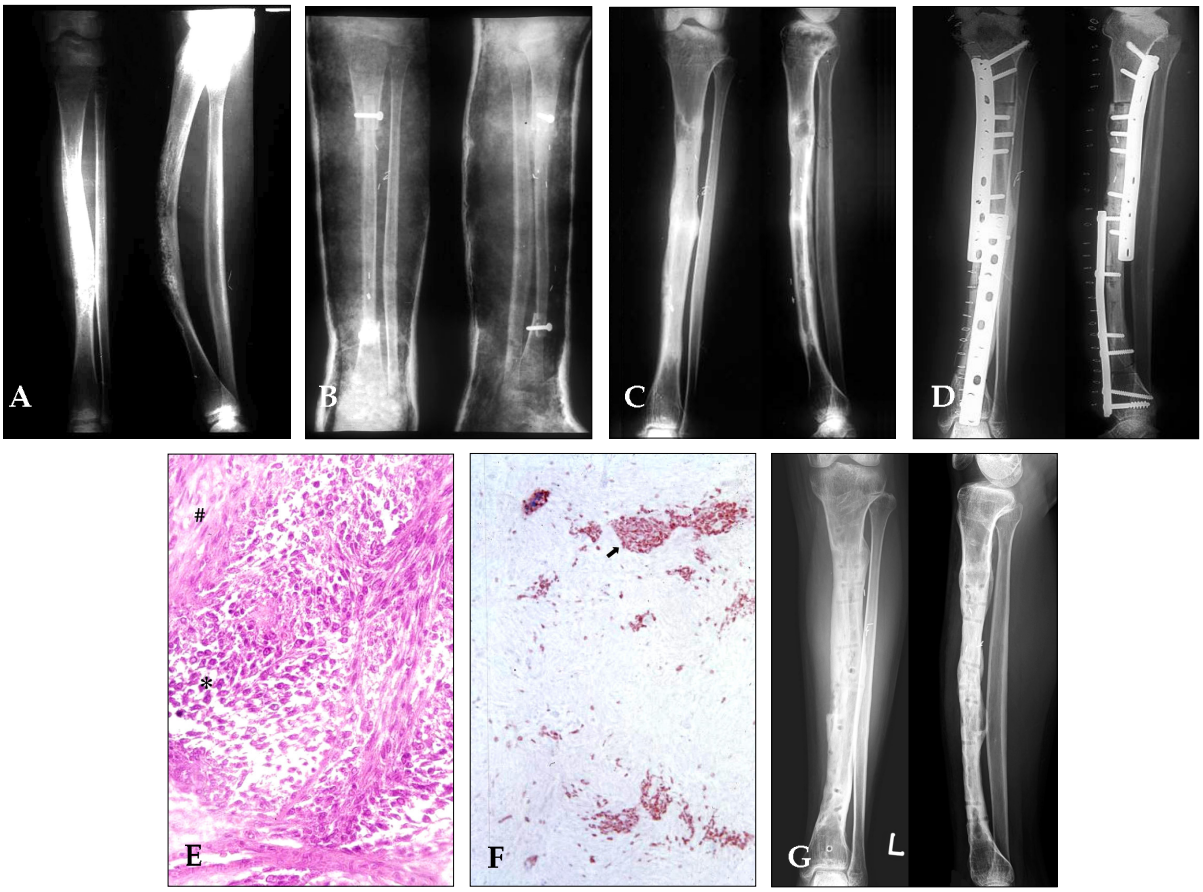

Fig. 1. (A) Initial radiographs, which were diagnosed as osteofibrous dysplasia, showed an extensive osteolytic lesion with a severe anterior bowing deformity of left tibia. (B) Radiographs after segmental resection and free vascularized fibular graft. (C) Postoperative 14 years, this patient had an osteolytic round lesion on the junction of the grafted fibula and tibia, thus suggesting a recurrence. (D) The recurrent lesion was excised and reattached after pasterization. (E) Photomicrographs representing the lesion area and showing the large nests of epithelial neoplastic cells in the dense sclerotic stromal tissue, which are compatible with classic AD (H&E × 200). (*; epithelial cells, #; stromal tissue) (F) Cells showing a positive immunohistochemical staining for cytokeratin (× 100). (G) The last follow-up radiograph showed no evidence of recurred lesion (4 years and 11 months after the second operation).

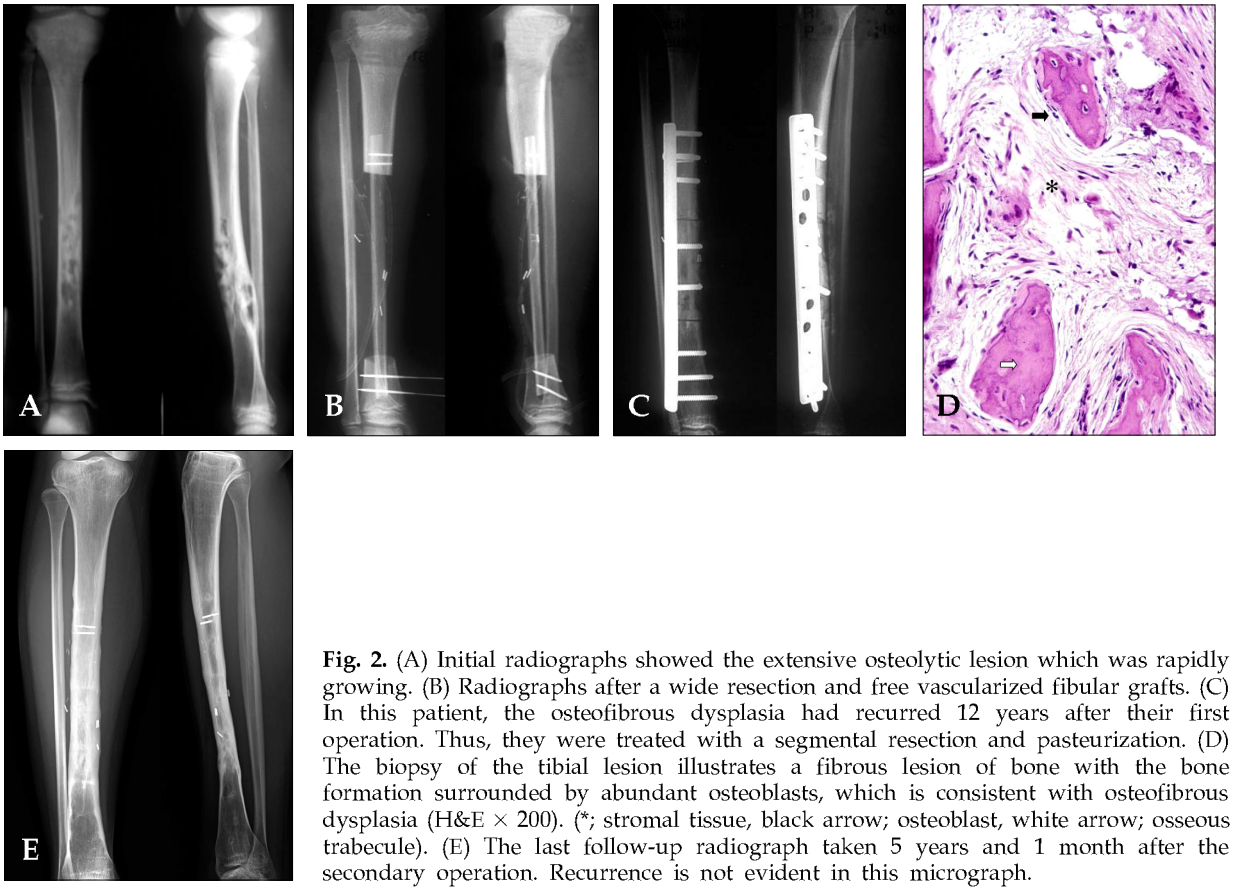

Fig. 2. (A) Initial radiographs showed the extensive osteolytic lesion which was rapidly growing. (B) Radiographs after a wide resection and free vascularized fibular grafts. (C) In this patient, the osteofibrous dysplasia had recurred 12 years after their first operation. Thus, they were treated with a segmental resection and pasteurization. (D) The biopsy of the tibial lesion illustrates a fibrous lesion of bone with the bone formation surrounded by abundant osteoblasts, which is consistent with osteofibrous dysplasia (H&E × 200). (*; stromal tissue, black arrow; osteoblast, white arrow; osseous trabecule). (E) The last follow-up radiograph taken 5 years and 1 month after the secondary operation. Recurrence is not evident in this micrograph.

Case 2

A girl aged 8 years and 2 months (case 4 in Table 1) presented with a known osteolytic lesion showing rapid progression in her right tibia. Under the diagnosis of OFD, a wide resection and reconstruction with free vascularized fibular bone grafts were performed (Fig. 2A and B). Twelve years after this operation, a resection and pasteurization were performed for the recurrent lesion (Fig. 2C). Review of the initial pathological slide showed no epithelial cells with the HE staining (Fig. 2D) and the immunohistochemical staining revealed few cytokeratin positive cells. These findings were concurrent with OFD. For 5 years and 1 month after the second operation, we found no evidence of recurrence and the patient showed a full range of motion in her knee and ankle joint (Fig. 2E).

Case 3

A 2-year-old boy presented with a palpable mass on his right proximal tibial area. A biopsy indicated a diagnosis of OFD. Since the lesion showed a rapid progression with a wide involvement of tibia, segmental resections, and reconstruction with osteogenesis was performed by an internal transport with Ilizarov external fixation apparatus. The lesion recurred 18 months after the surgery and subperiosteal curettage and bone grafts were performed on the recurrent lesion. No evidence of recurrence was found for 11 years after the second operation. On reviewing initial and recurrent biopsy slides, a few nests of epithelial cells within the fibrous stroma rendered a diagnosis to OFD-like AD, which stained positive for CK.

DISCUSSION

OFD is a rare, benign bone tumor that occurs mainly in the tibia and consists of a 0.2% primary bone tumor.[1,8] These findings are also reported in

Korea by several authors including Hahn et al.[17] The tumor is known to affect children under age 20, before the termination of bone growth.[3,9,18] The tumor is more frequently found in males. Our study had more female patients, and 19 out of 20 of the cases occurred before the age of 20. The one remaining case was a 65-year-old man with a 20 mm round cortical mass in his proximal tibial shaft area, which was removed with an en bloc excision. The pathology showed OFD findings, but required differentiation with an osteoblastoma. In all cases of patients over 15 years old, close observation was preferentially performed after a confirmation of OFD by biopsy, and all showed a benign course.

Several follow-up data showed the spontaneous regression of OFD after puberty.[9,10,19] Campanacci and Laus recommended not to perform subperiosteal curettage in OFD patients younger than 15 years old because they recur in most cases.[9] In addition, they also recommended managing cases by a conservative method, even in cases of pathological fractures. However, in cases of bony weakness due to wide involvement, rapid progression, or recurrent pathological fractures, the recommended method is surgical treatment with wide resections and bony reconstructions.[1,4,9,18] Campanacci et al and Nakashima et al reported that the recurrence rate after curettage was between 64-100%.[9,20] According to Kempson, removal of the periosteum around the lesion is important to prevent recurrence, and therefore, subperiosteal curettage or subperiosteal excision is not appropriate.[21] Some authors claim that early surgical intervention should be more radical than the previously recommended interventions.[22,23]

In our study, 2 cases treated with subperiosteal curettage did not recur, while all 5 cases treated by wide resection and bony reconstruction required a second operation for the recurrent lesion. All 6 cases were managed with conservative treatment after the biopsy because the lesion was small and showed a benign course; therefore, no additional operation was necessary. These results show that if the initial lesion has a large involvement or rapid progression, a wide resection cannot prevent recurrence.

The most important factors related to disease recurrence or progression are patient age at the time of diagnosis, lesion size, and the degree of initial disease progression.[3,18,19] In this study, all cases of patients over 15 years old were allowed to undergo conservative management because all of these patients showed stable progress. But recurrences were more frequent even after a wide resection of the lesion in the patients under 15 years old with large or rapidly progressing tumors. Therefore, maintaining a conservative or non-aggressive management is recommended for patients under 15 years old. A more aggressive treatment is recommended for patients over 15 years because it is helpful in lowering the recurrence rates. However, in the cases of rapid disease progression with involvement of the entire tibia or in cases of recurrent pathological fractures, a wide resection followed by reconstruction is necessary. And the severe bowing deformity also requires an early correctional operation.[1,3,4,9,17,18,22,23]

The diagnosis of OFD requires differentiation from fibrous dysplasia and AD.[5,15,24] The similarity of OFD to fibrous dysplasia had been previously proposed, but fibrous dysplasia and OFD can be histopathologically differentiated by looking for the presence of osseous tissue. However, similar clinical characteristics, radiological findings, as well as pathological findings sometimes make differentiating OFD from AD very difficult. Many hypotheses have been proposed on the relationship between OFD and AD. Since Dockerty and Meyerding first presented the overlap of the two diseases in 1942,[25] some reports have pointed out the relativeness of OFD and AD.[1-7,9,12-16,26] One hypothesis suggested that OFD is a precursor of AD.[1-4] Another theory proposed that OFD is a secondary disease formed in the process of degeneration or removal of the AD tumor cells.[5-7] Conervative treatment and excision after the cessation of growth is the principle behind OFD treatment, whereas radical resection or amputation is necessary for AD.[9-11] Therefore, if OFD is a precursor for AD, then early radical resections could be necessary.

OFD usually occurs in patients before the end of growth and originates from the tibial cortex. According to Campanacci and Laus, AD is very rare before the age of 10,[9] but Hazelbag et al reported 4 AD cases in patients under 10 years old in a study including a total of 32 ADs.[13]

Immunohistochemical staining showing scattered, isolated CK positive cells within the fibrous stroma of OFD indicates the histogenetic relationship between OFD and AD.[2,6,10,12,16] In addition to the clinical, radiological, and histological findings, the common histogenesis of OFD and AD are also suggested by immunohistochemical and cytogenetic studies.[4,27] Cytokeratin reacts positively in tumor cells that originate from epithelial cells. According to a study on the antibodies against a subtype of cytokeratin by Hazelbag et al, cytokeratin (CK) in AD is different from synovial sarcoma, chordoma, or epithelial sarcoma.[13] Benassi et al. insisted on a correlation between OFD and AD based on their results that showed positive CK19 and negative CK8 and CK18 staining in both OFD and AD cases.[12] In a cytogenetic study, both OFD and AD have trisomies on chromosomes 7, 8, and 12.[4,16]

Czerniak et al. first named the mixture of classical AD and OFD as differentiated AD.[5] Currently, this disease state is called OFD-like AD. OFD-like AD develops in younger patients than classical AD. Patients with OFD-like AD have small nests (< 25 cells) or strands of epithelial cells within the fibrosseous stroma that can be observed with HE staining. OFD and OFD-like AD are very similar in other physical features that can be visualized by HE staining. Many authors are reporting difficulty in differentiating the histological features of OFD, OFD-like AD, and AD.[2,4,5,12-16] Currently, AD is diagnosed when the epithelial cells form large nests, whereas OFD-like AD is diagnosed if these nests are relatively small. However, in many cases, classical AD has some stromal component with a OFD like appearance. Therefore, even after OFD is diagnosed by incisional biopsy, AD or OFD-like AD could possibly occur in other regions within the same patient.

Springfield et al diagnosed 6 out of 10 cases of recurrent OFD as AD, and insisted that OFD is the precursor of AD.[15] Some case reports have recorded a change of the initial OFD diagnosis to AD in the recurrent lesion. In some cases, reviewing the initial slide generated from the first biopsy caused the clinician to change the diagnosis to AD.[13,15] Explanations for these changes could be either inadequate specimen sampling from the initial biopsy or an incorrect pathological reading. However, one case report has noted a case where after the complete excision of the initial lesion, OFD changed into classic AD via an OFD-like AD stage.[13,15] Another case reported OFD-like AD with metastasis to the lung.[28]

By taking these reports into account, OFD is sometimes regarded as a precursor of AD, while OFD-like AD is considered to be the intermediate form of the disease.

Therefore, a diagnosis of OFD always requires attention, because the disease could actually also have AD in accompanying findings or OFD could differentiate into AD.

In our study, a patient developed AD 14 years after the initial treatment of OFD. The patient received an early wide resection and reconstruction with a free vascularized fibular bone graft after pathologic confirmation of OFD. Fourteen years after the operation, the osteolytic lesion recurred at the proximal margin of grafted fibula and remaining tibia. Biopsy of the recurrent lesion showed the typical findings of AD. For a more accurate diagnosis, cytokeratin immunohistochemical staining was performed. After reviewing the initial pathological slide, the original diagnosis was compatible with OFD-like AD containing some scattered cytokeratin positive cells. The disease could have been initially diagnosed as OFD-like AD and then progressed to classic AD in the recurrent lesion.

After this case, we conducted a pathological review of all primary and recurred OFD cases in the same hospital and found another case that could be considered OFD-like AD. The original pathological diagnosis of the primary and recurred lesion for this particular case was changed from OFD to OFD-like AD.

Campanacci and Laus insisted on the possibility of giving a definite diagnosis of OFD without a biopsy if the radiological findings are typical and the location of the lesion as well as the age of patient are considered.[9] However, since OFD could possibly be the precursor to AD, a biopsy is mandatory. Although AD is radiologically very similar to OFD, AD must be pathologically differentiated from OFD because AD is a malignant tumor that spreads to other organs and has a long clinical course. Therefore, a biopsy and evaluation of the epitheloid component must be performed to

differentiate between OFD and AD. Furthermore, if possible, the confirmation of cytokeratin positive cells by immunohistochemical analysis is also needed for these patients.

Patients diagnosed with OFD require close observation because the disease is often locally aggressive, and could actually be AD or OFD-like AD. Since early radical resection could not prevent recurrence, an advisable course of treatment is to administer a less aggressive treatment until the growth ceases.

An OFD diagnosis requires very special attention because similar radiological and pathological characteristics are shared between OFD, OFD-like AD and classic AD. In addition, patients with OFD could possibly convert from OFD to OFD-like AD or classic AD. Furthermore, a work up for metastasis is needed for patients with OFD-like AD or classic AD because these disease types are capable of metastasis. Immunohistochemical staining with cytokeratin may be helpful in differentiating between OFD and AD.

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