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Case report

A patient with bizarre parosteal osteochondromatous proliferation in the mandible

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ABSTRACT

Bizarre parosteal osteochondromatous proliferation lesions in the head and neck region are very rare. Clinically, these lesions present as protrusions of bone, usually in the small bones of the hands and feet. This report is of a 9-year-old girl with a bizarre parosteal osteochondromatous proliferation lesion in the mandibular mental region, a site that has not been described previously. Based on the physical examination and X-ray analysis, mandibular osteoma was clinically diagnosed. The tumor was locally excised and pathologically diagnosed as bizarre parosteal osteochondromatous proliferation. Five months later, the lesion recurred in the same region and was again locally excised. Postoperative recovery was uneventful and there has been no recurrence since the second operation, more than 8 years ago.

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1. Introduction

Bizarre parosteal osteochondromatous proliferation (BPOP) is a rare benign lesion usually involving the small bones of the hands and feet as proposed by Nora et al. in 1983 [1]. However, the present patient developed a lesion in the mandibular mental region. It appears extremely rare for BPOP to present in this location, and to the best of our knowledge, the literature contains no similar cases. Studies have shown no gender differences in the clinical distribution of BPOP. In one series, age at onset ranged from 8 to 73 years [2], whereas another reported that age of onset ranged from 14 to 74 and that the peak age of onset was 25 to 35 [1]. Previous research has not produced any definitive conclusion as to the ideal timing for removal of these lesions, and the optimal method of excision remains unclear [3].

2. Case report

The patient, a 9-year-old girl, initially consulted our hospital in June 2000, after developing swelling in the mental area of the mandibular region. She had no relevant personal medical history or family history. In August 1999, a protrusion was noticed in the mental region, but no medical attention was sought. From April 2000, the swelling rapidly increased in size and the patient was conse-

quently taken to a local internal medicine clinic and then referred to our department.

Examination of the extraoral right submental region revealed a hard, bony protrusion. No other abnormal findings were noted (Fig. 1). Intraoral examination showed nothing unusual. X-ray showed a protrusion from the mandibular cortical bone. There was no continuity between the protrusion and the mandibular body or marrow cavity (Fig. 2). Based on the physical examination and X-ray analysis, mandibular osteoma was clinically diagnosed.

Intraoral local excision was performed under general anaesthesia. A bony protrusion from the mandible mental region measuring approximately 3 mm was noted. The lesion had no adhesions to the periosteal tissue, which facilitated exfoliation, and the bone surface was smooth. The periosteum was exfoliated and the protruding bone and about 2 mm of the peripheral cortical bone were resected. Finally the exfoliated periosteum was placed back into the resected area. A detailed histopathological examination was performed and showed outgrowths of bone, juvenile cartilage, and fibroblasts. The nuclei of the osteocytes and fibroblasts demonstrated no atypia, but those of the chondrocytes showed slight atypia in the form of anisokaryosis and irregular shape (Fig. 3). Based on these findings, a histopathological diagnosis of BPOP was made.

The postoperative course was favourable, but swelling of the right submental region was once again observed in January 2001. Plain X-ray and computed tomography again revealed a well-demarcated bony protrusion, measuring approximately 3 mm and extending from the mandibular cortical bone (Fig. 4). As before,

Abbreviation: BPOP, bizarre parosteal osteochondromatous proliferation.

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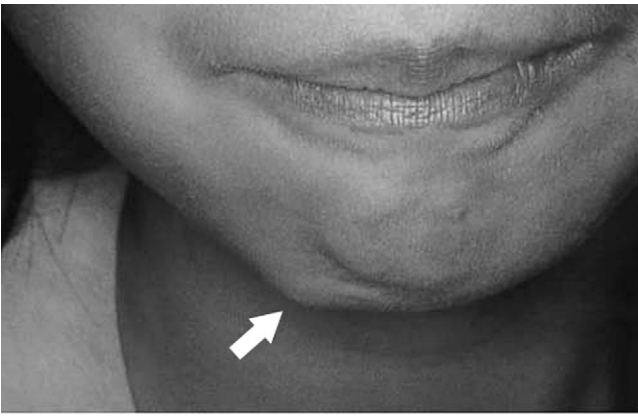


Fig. 1. Preoperative view. Swelling is evident in the right submental region (arrow).



Fig. 2. X-ray appearance. A protrusion from the mandibular cortical bone (arrow) can be seen.

there was no continuity with the marrow cavity. These findings suggested recurrent BPOP.

In April 2001, intraoral local excision was performed under general anaesthesia (Fig. 5). Posterior to the previous surgical site, a new bony protrusion was found connected to the mandibular cortical bone and measuring approximately 3 mm. At the previous surgical site, bone regeneration was observed. Granulation tissue was also noted in a portion of the periosteum adjacent to the end of the new bony protrusion; we therefore resected the new pro-

trusion, the granulation tissue, all of the periosteum covering the lesion and 4 mm of the peripheral cortical bone from both sites. As of May 2008, the course of recovery has been favourable without any sign of recurrence or facial deformity (Fig. 6).

Histopathological diagnosis from the second surgery showed outgrowths of cartilage, osteoid tissue, as well as fibroblasts in the protrusion site. Slight atypia was noted in the hyaline cartilage. As

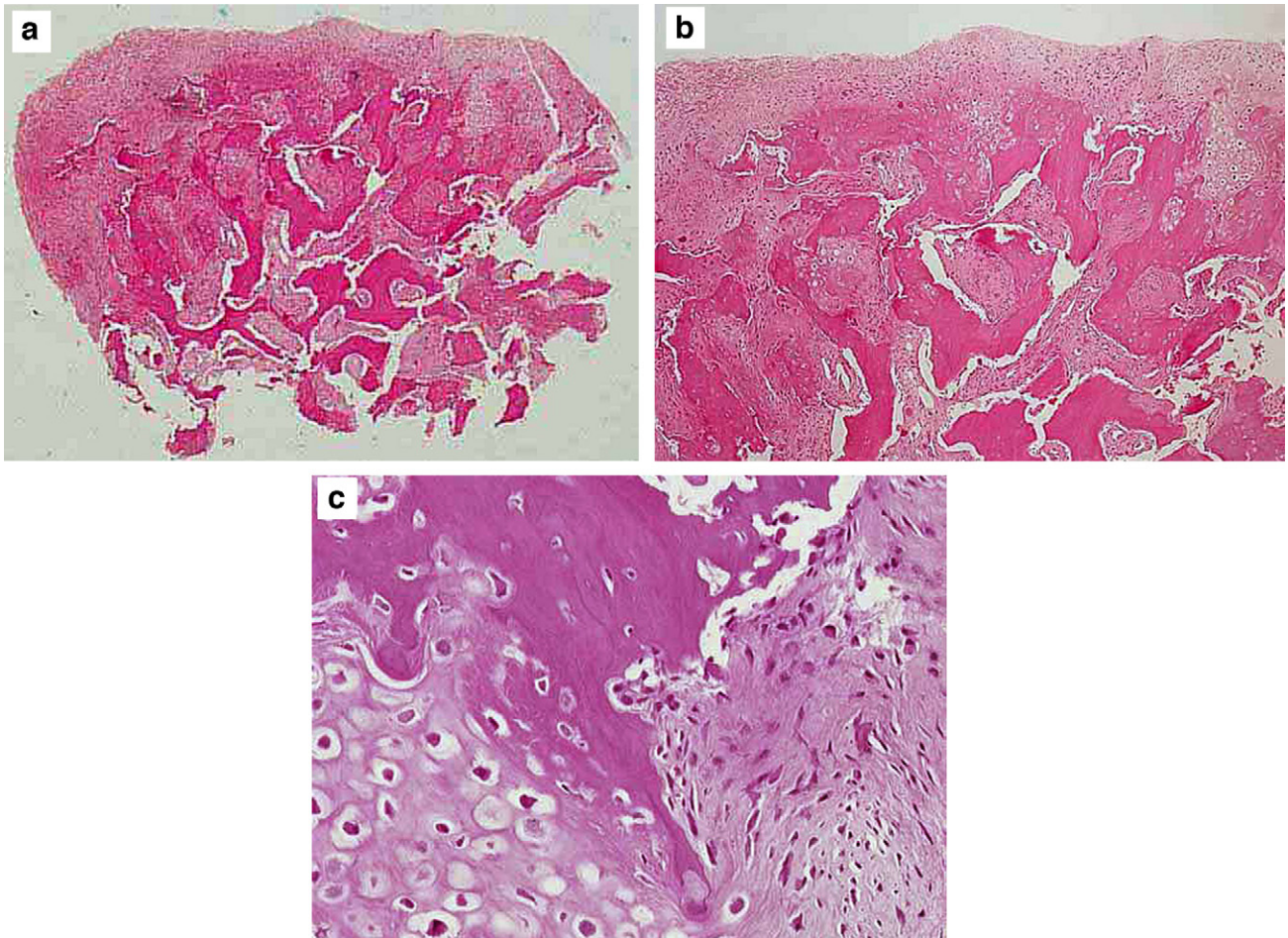


Fig. 3. Histopathological findings at initial surgery. (a) Outgrowths of bone are visible (hematoxylin and eosin; original magnification 20 \times). (b) Layer showing cartilage and osteoid structure (hematoxylin and eosin; original magnification 40 \times). (c) Slight atypia in the form of anisokaryosis and irregular shape are noted in the nuclei of the chondrocytes (hematoxylin and eosin; original magnification, 200 \times).

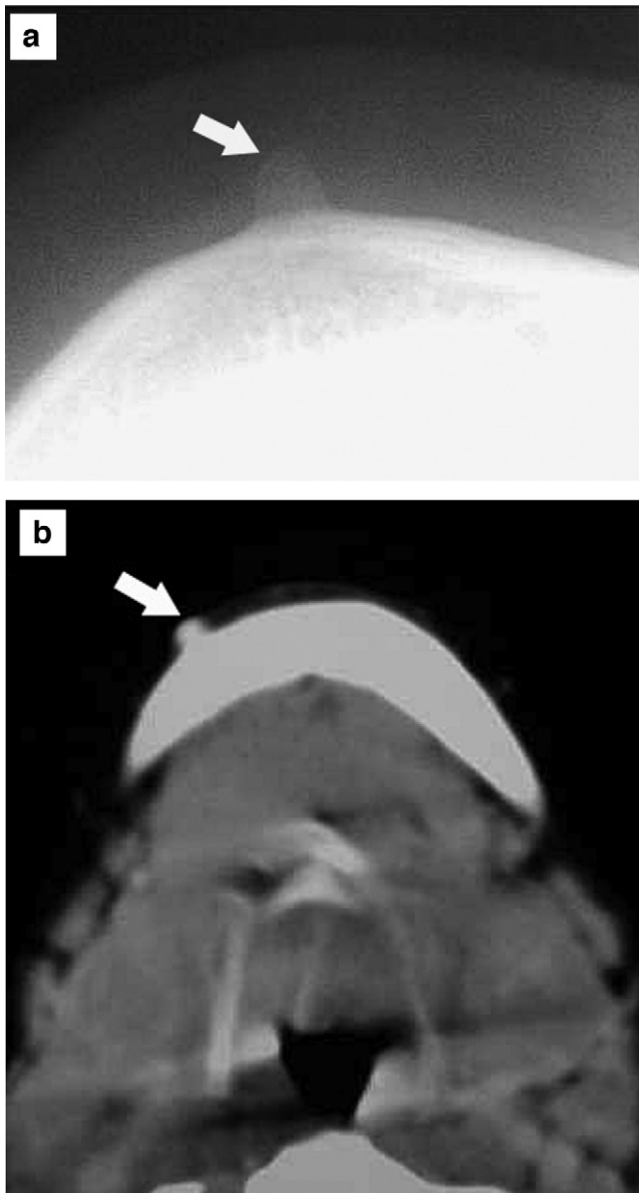


Fig. 4. Plain X-ray and axial computed tomography images. Plain X-ray (a), and axial computed tomography (b) showing a protrusion approximately 3 mm in length (arrow).

described in the histopathological findings from the initial surgery, neither the osteocytes nor fibroblasts showed atypia. The peripheral bone was well differentiated, and there was no infiltration of atypical cells into the thickened periosteum (Fig. 7). A comparative analysis of the histopathological findings from both surgeries showed that, compared with the findings after the first surgery, the second specimen demonstrated a thickening of the trabecular bone, homogenisation of the bone matrix, bone cell shrinkage, and fewer osteoblasts. Binucleated chondrocytes with bizarre morphology were also present (Fig. 8). Furthermore, the fibroblast content in the stroma had greatly decreased, while the collagen content had considerably increased. These findings indicate bone maturation rather than malignancy.

3. Discussion

BPOP is a rare, idiopathic, non-neoplastic lesion, and is also considered to be a reactive lesion, as are florid reactive periostitis

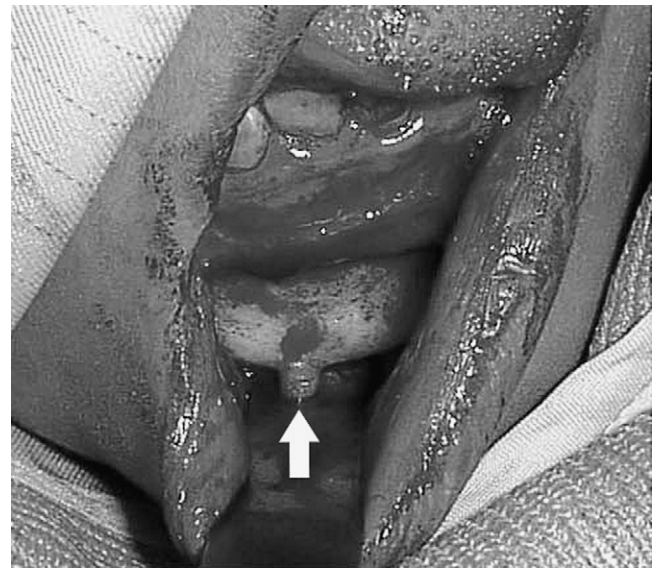


Fig. 5. Operation view. A 3-mm bony protrusion connected to the mandibular cortical bone was detected posterior to the area resected during the previous surgery (arrow).

and myositis ossificans [1,2]. However, the process of growth and retraction remains to be clarified. In the English literature, studies have reported no gender difference in the incidence [2], that the age at onset ranged from 8 to 73 years [2] and 14 to 74 years [1] and that the most common age range at diagnosis was 20 to 30 years [2] and 25 and 35 years [1]. The sites most commonly reported include the short bones of the hands, the short bones of the feet and the long bones [2,4]. Reports of BPOP in the head and neck region

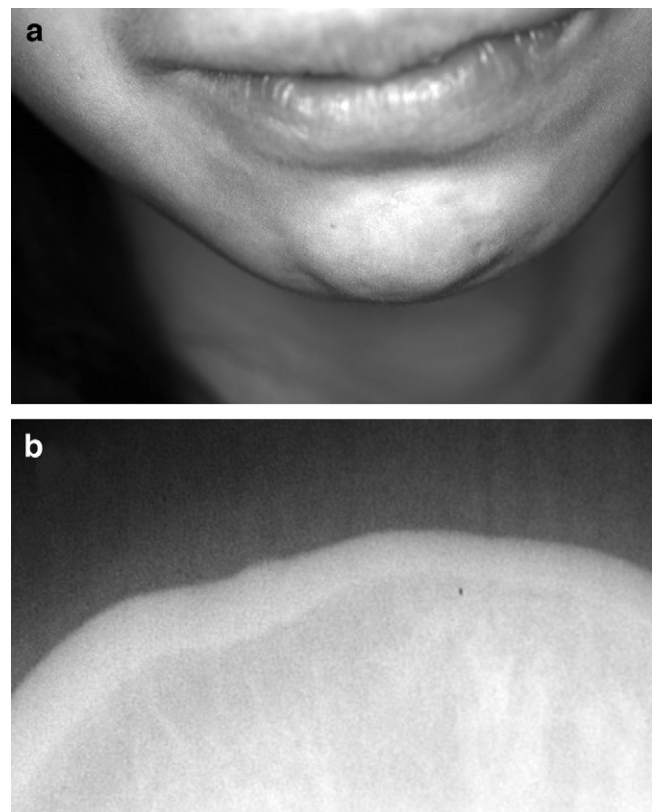


Fig. 6. Postoperative view. (a), (b). The course of recovery has been favourable without any sign of recurrence or facial deformity.

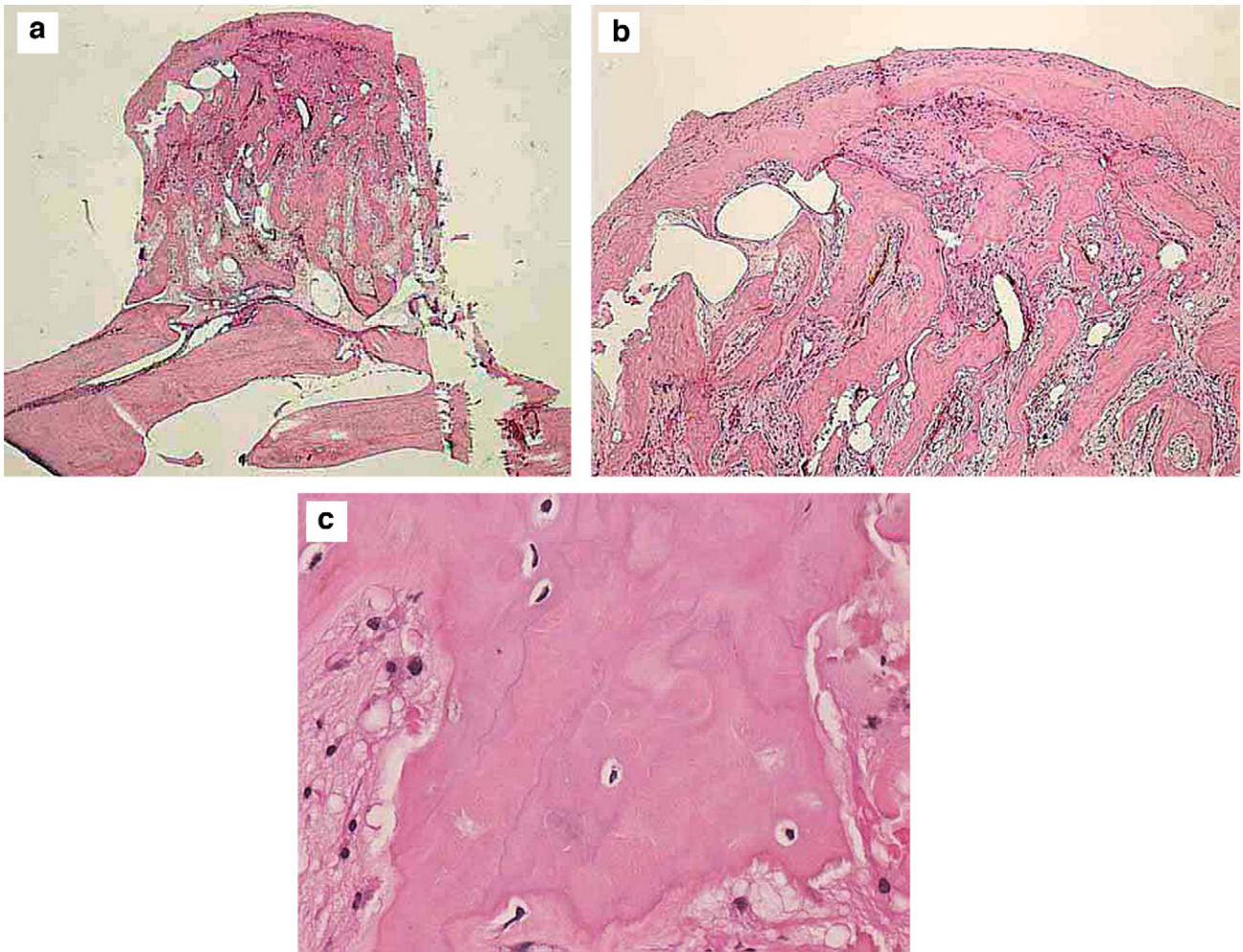


Fig. 7. Histopathological findings from the second surgery. (a) Outgrowths of cartilage are seen (hematoxylin and eosin; original magnification 20 \times). (b) Layer showing cartilage and immature bone structure (hematoxylin and eosin; original magnification 40 \times). (c) Osteoid shows no atypical features (hematoxylin and eosin; original magnification 400 \times).

are extremely rare, with only one case report involving the upper anterior teeth in the maxillofacial region [2,4].

In the present study, BPOP developed in the mandibular mental region of the maxillofacial area, a location that has not previously been reported.

According to several studies, one-third of patients had a history of physical trauma, and the interval until onset ranged from 2 months to a few years. However, as in the present case, many patients report no such history [2].

The differential diagnosis of BPOP comprises two categories: one includes benign reactive lesions, benign bone tumours of the cartilage system such as osteochondroma, subungual exostosis, florid reactive periostitis, and ossification-related myositis. The other includes extrasosseous sarcomas such as parosteal osteosarcoma, parosteal chondrosarcoma, chondrosarcoma, and periosteal chondrosarcoma. However, BPOP has specific radiological and histopathological features and can be differentiated from other types of osteochondroma-related proliferations [1,2].

In terms of radiological features, BPOP is visualised as a bony lesion with a clear border, protruding from the bone surface; i.e., similar to osteochondroma [1,2]. However, unlike osteochondroma, BPOP is associated with no or very slight changes in the bone cortex. Furthermore, BPOP demonstrates no continuity between the lesion and marrow cavity, and no abnormalities in the bone

marrow. BPOP lesions have a non-specific presentation on magnetic resonance imaging [5].

In the present patient, a bony protrusion with a clear border was detected in the mandibular cortical bone. There were no changes in the bone cortex and no continuity with the marrow cavity. These features all pointed towards a diagnosis of BPOP.

Histopathological features of BPOP include proliferative changes in bone and cartilage, and spindle-shaped fibroblasts, as are common in florid reactive periostitis [1,2]. However, in patients with BPOP, bone/cartilage formation is more marked. A cartilage cap is usually observed, and irregular maturation or ossification of the bone tissue immediately below this is noted. In BPOP, the cellular density of cartilage is high, and relatively large chondrocytes are present. These cells include binuclear and atypical cells which may suggest malignancy [1,2]. Key indicators of parosteal osteosarcoma are atypical osteoblasts and fibroblasts as well as infiltration into the peripheral tissue [1,2]. Whereas, in the case of BPOP, nuclear staining with chromatin does not show any dark areas, and neither atypia of the fibroblasts nor infiltration into the peripheral tissue is usually observed. Bone tissue is formed from interstitial spindle cells, and cartilage in the bone area is stained blue [1,2]. In another study, Dorfman described a spectrum of reactive lesions of which the first stage is known as florid reactive periostitis and pathologically consists of spindle cells with minimal osteocartilaginous proliferation. The second stage characteristically shows new

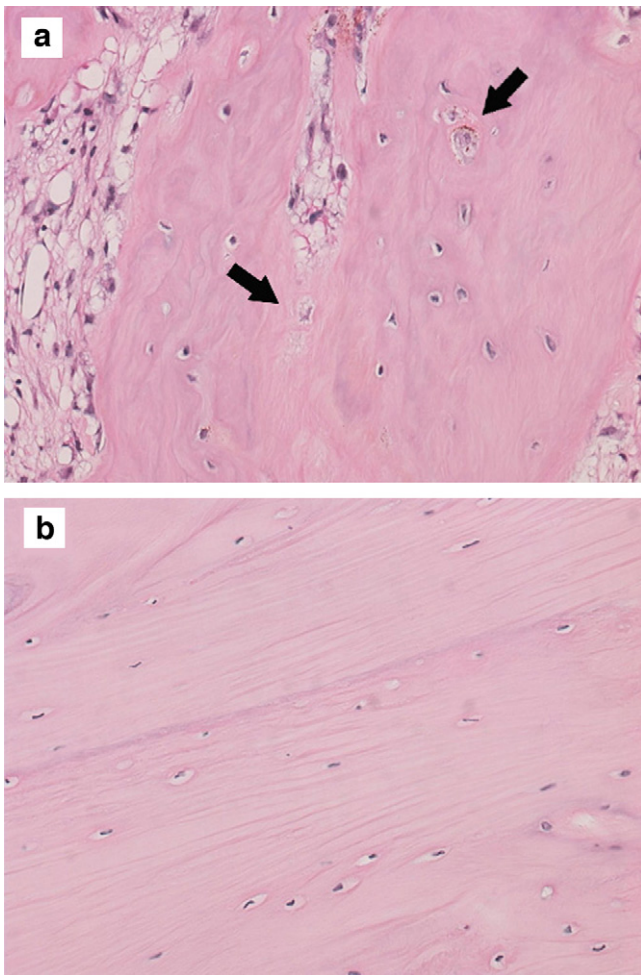


Fig. 8. Histopathological findings from the second surgery. (a) Outgrowths of cartilage are seen and binucleate and bizarre chondrocytes were also present (arrows) (hematoxylin and eosin; original magnification 400 \times). (b) Lamellar bone (hematoxylin and eosin; original magnification 400 \times).

bone and metaplastic cartilage that becomes more prominent (i.e., BPOP), and is composed of bizarre cartilage with hypercellularity and binucleated cells, blending together with woven bone against a fibrous background. In the third stage, the focus of ossification matures and a bony base is formed with a cartilage cap [6].

In the present patient, outgrowths of osteoid tissue, juvenile cartilage and fibroblasts were noted. Although there were no atypical osteocytes or fibroblasts, slight atypia resembling anisokaryosis and irregular nuclear shape were observed in the chondrocyte nuclei. The peripheral bone tissue was normal and there was no infiltration of atypical cells into the thickened periosteum. Furthermore, a comparative analysis of the histopathological findings from the initial and second surgeries showed thickening of the trabecular bone, homogenisation of the bone matrix, bone cell shrinkage, and a decrease in the number of osteoblasts. Moreover,

the stroma exhibited a large decrease in fibroblast content and a considerable increase in collagen content. These findings indicated bone maturation rather than malignancy. Considering all of these histopathological findings, we ruled out fibrous dysplasia, ossifying fibroma, and parosteal osteosarcoma, and made a diagnosis of BPOP.

Although there have been no reported cases of metastatic BPOP, the rate of recurrence is relatively high [1,2]. Nora et al. reported that the recurrence rate after 2 months to 2 years was 51%, and that 22% of those patients had a second recurrence [1]. Meneses et al. found a similar incidence of recurrence [2].

The ideal timing for removal of these lesions has not been defined; however, Yuen et al. suggested that recurrence is likely when excision is carried out too early or the periosteum is not fixed (i.e., florid reactive periostitis). Furthermore, they stated that recurrence is more frequent in the exophytic type of completely ossified lesions (BPOP) [7].

Although, the optimal method of excision also remains unclear [3]; previous research tends to promote local excision, even in the case of recurrence, as the best option for removal [4,8,9].

In the present case, relapse occurred 6 months after the initial surgery despite resection of the lesion and 2 mm of the peripheral cortical bone. However, in the second surgery, the lesion together with 4 mm of the peripheral cortical bone was resected, resulting in no subsequent recurrence. Therefore, in the case of recurrence, it would seem that local excision with slightly wider margins should also be considered as a potentially successful treatment option.

One very important point to consider when selecting either treatment option is whether it will lead to a facial deformity. In the present case, two operations were performed, with the second surgery involving local excision of the actual lesion with slightly wider margins. Considering that there has been no additional recurrence or facial deformity in our patient for more than 8 years after the second surgery, the treatment selected in our second operation may be considered a good option. Irrespective of which course of treatment is selected, long-term follow-up must be continued because of the possibility of recurrence.

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