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CASE REPORT

Actinomycotic osteomyelitis with proliferative periostitis arising in the mandibular ramus: an unusual case with spontaneous bone regeneration after coronoidectomy

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Abstract

Actinomycotic osteomyelitis is an aggressive and persistent disease capable of invading and destroying bone, and chronic osteomyelitis with proliferative periostitis represents new bone formation with periosteal reaction. We report a rare case of actinomycotic osteomyelitis with proliferative periostitis arising in the mandibular ramus and spontaneous bone regeneration after coronoidectomy. A 14-year-old girl was referred for swelling in the right parotid-masseteric region and severe trismus. Contrast-enhanced CT revealed that heterogenous enhancement of the right masseter muscle, and a reactive bone formation over the lateral cortex of the right mandibular ramus and osteolysis of the condyle were seen in plain CT. MRI showed that the mandibular ramus was a low-signal intensity and the reactive bone on the ramus was signal intensity similar to muscle on T1-weighted images. The lesion was clinically and radiologically diagnosed as chronic osteomyelitis of the mandibular ramus. However, a biopsy was performed intraorally under general anesthesia to rule out a malignant bone tumor, and pathological examination showed fibrous bone and *Actinomyces* druses. Finally, the lesion was diagnosed as actinomycotic osteomyelitis with proliferative periostitis. She underwent image-guided intraoral removal of impacted right third molar and reactive proliferative bone on the right mandibular ramus under general anesthesia. To improve trismus, coronoidectomy also was performed. After the discharge, AMPC was administrated intraorally for 7.5 months. Postoperative panoramic radiograph and CT showed the right mandibular angle resorption and coronoid process regeneration. There was no recurrence of mandibular osteomyelitis 7 years after surgery.

Keywords Actinomycotic osteomyelitis · Malignant bone tumor · Mandible · Proliferative periostitis · Spontaneous bone regeneration

Introduction

Actinomycosis is a specific chronic infection caused by *Actinomyces* species, which are anaerobic, Gram-positive, and filamentous bacteria [1, 2]. *Actinomyces* are normal inhabitants of the oral cavity and typically have low pathogenicity, but become pathologic when gaining access to the subcutaneous tissues. Cervicofacial actinomycosis is most common among three forms (cervicofacial, pulmonary or pulmothoracic, and abdominal–pelvic actinomycosis) and accounts for more than half of all reported cases [1, 3]. Of patients with cervicofacial actinomycosis, 11.7–15% had bone infection such as periostitis and osteomyelitis [1, 3]. Actinomycosis is an aggressive and persistent disease capable of invading and destroying bone, although bone involvement is not seen until the later stages of the disease [4].

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As an even rarer case, pathological mandibular fracture is caused by actinomycotic osteomyelitis [5].

Chronic osteomyelitis with proliferative periostitis is a rare disease, and represents new bone formation with periosteal reaction [6]. This osteomyelitis has been described under different terms, such as Garré's osteomyelitis, proliferative periostitis, and periostitis ossificans [6, 7]. This type of osteomyelitis is more common in younger patients, and the lesions most prevalent areas are the premolar and molar areas of the mandible [6, 8]. Sources of infection of the jaw include dental caries associated with periapical periodontitis, periodontitis, fracture, and nonodontogenic infection [6, 8]. We report a rare case of actinomycotic osteomyelitis with proliferative periostitis arising in the mandibular ramus and spontaneous bone regeneration after coronoidectomy.

Case report

A 14-year-old girl visited a private dental clinic for trismus (maximum mouth opening: 20 mm) and pain in the right temporomandibular region. Although conservative treatment was performed for 3 months after the diagnosis of temporomandibular disorder, the symptoms were not improved. Because swelling in the right parotid-masseteric region and further severe trismus occurred (Fig. 1), she was referred to our department. The maximum mouth opening was 10 mm, and there was no right mental nerve palsy. There was no history of a previous nonspecific infection or trauma. Intraoral examination showed gingival swelling in the right retromolar region. Hematological examination showed a white blood cell (WBC) count of 7400/μL and C-reactive protein (CRP) level of 0.3 mg/dL. Panoramic radiograph showed that radiopaque appearance of the right mandibular ramus and impacted mandibular third molar without root formation in the right mandibular ramus (Fig. 2). Contrast-enhanced computed tomography (CT) revealed that heterogenous

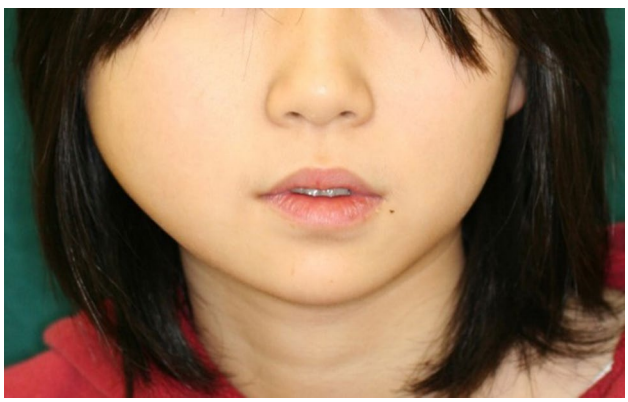

Fig. 1 Facial view shows swelling in the right parotid-masseteric region

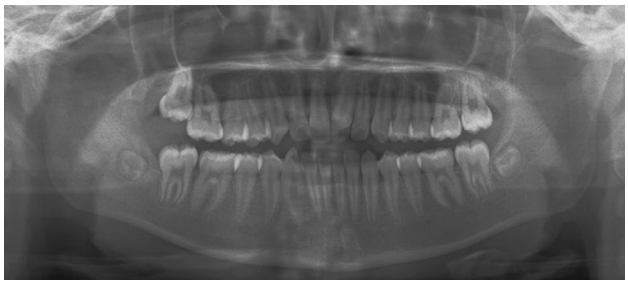

Fig. 2 Panoramic radiograph shows that radiopaque appearance of the right mandibular ramus and impacted mandibular third molar without root formation in the right mandibular ramus

enhancement of the right masseter muscle, and a reactive bone formation over the lateral cortex of the right mandibular ramus and osteolysis of the condyle were seen in plain CT (Fig. 3). Magnetic resonance imaging (MRI) showed that the mandibular ramus was a low-signal intensity and the reactive bone on the ramus was signal intensity similar to muscle on T1-weight images (Fig. 4). Gadolinium-enhanced T1-weighted images and short TI inversion recovery (STIR) images revealed heterogeneous, intermediate-to-high-signal intensities of the right masseter muscle and the reactive bone. Fat-suppressed gadolinium-enhanced T1-weighted images showed heterogeneous high-signal intensity of the right masseter muscle and the reactive bone. In 2-deoxy-2-[^{18}F] fluoro-D-glucose (FDG) positron emission tomography, FDG uptake of the right mandibular ramus was high (maximum standardized uptake values = 4.4). The lesion was clinically and radiologically diagnosed as chronic osteomyelitis of the mandibular ramus. However, a biopsy was performed intraorally under general anesthesia to rule out a malignant bone tumor, and pathological examination showed fibrous bone and *Actinomyces* druses (Fig. 5). Finally, the lesion was diagnosed as actinomycotic osteomyelitis with proliferative periostitis. Ampicillin (ABPC, 2 g/day) was administrated intravenously for 10 days. After amoxicillin (AMPC, 750 mg/day) had been administrated intraorally, since the discharge from the hospital, postoperative right mandibular swelling decreased. Because the right mandibular swelling was recurrent 1.5 months after the biopsy and severe trismus remained, ABPC (2 g/day) was administrated intravenously for 2 weeks. As a radical treatment, intraoral removal of impacted right third molar and reactive proliferative bone on the right mandibular ramus under navigational guidance was scheduled. A resin occlusal splint with titanium markers and handle was manufactured to fix the reference frame to the mandible. CT was performed preoperatively after the occlusal splint was fixed with the patient's mandibular teeth, and digital imaging and communication in medicine (DICOM) data of the CT were transferred into a navigation system (Stealth Station®, Medtronic Inc.,

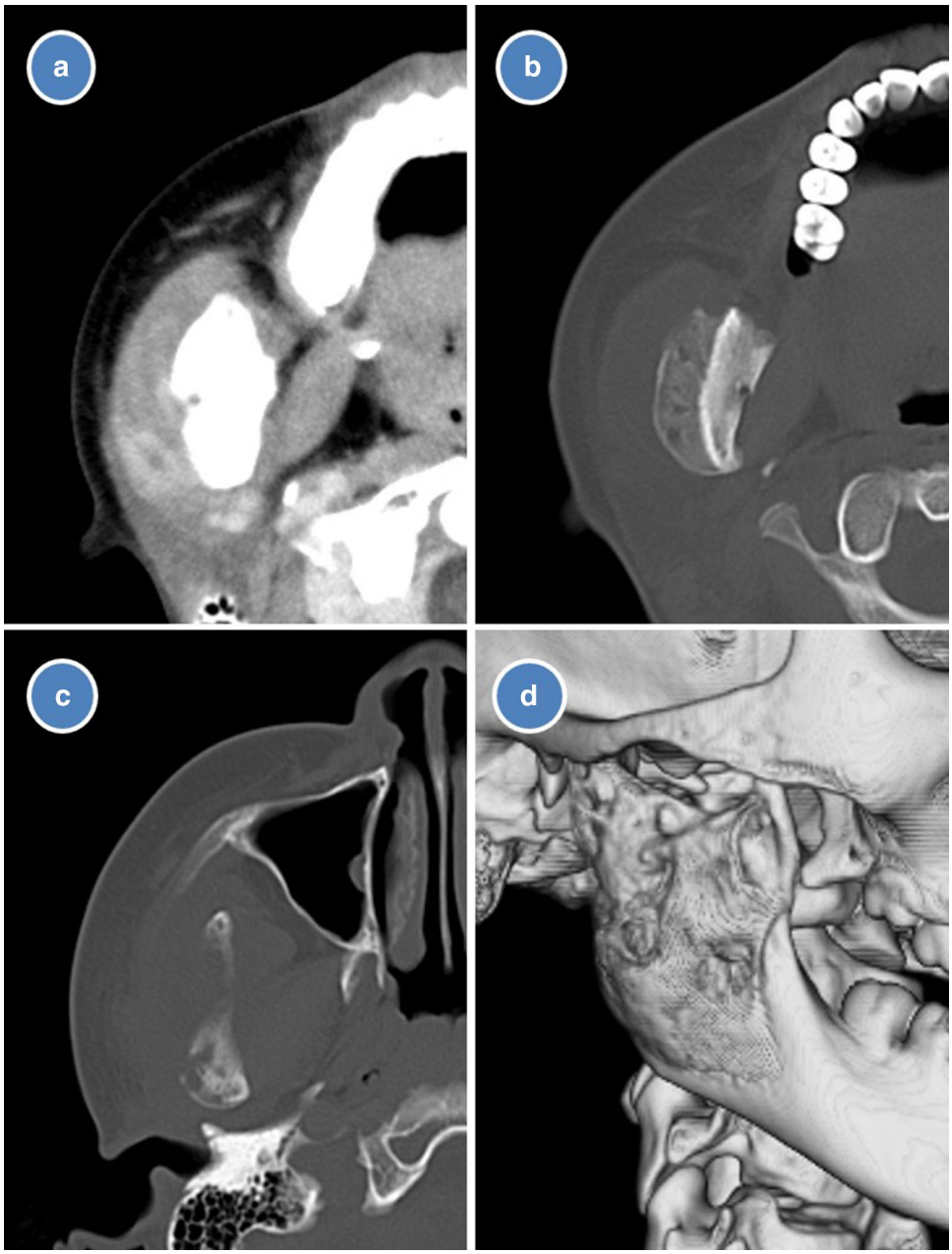

Fig. 3 Contrast-enhanced CT reveals that heterogenous enhancement of the right masseter muscle, and a reactive bone formation including multiple cystic lesions over the lateral cortex of the right mandibular ramus and osteolysis of the condyle are seen in plain CT. **a** Contrast-enhanced axial CT image, **b** and **c** Axial CT in bone window, and **d** 3D-CT image

MN, USA). The patient underwent image-guided intraoral removal of impacted right third molar and reactive proliferative bone on the right mandibular ramus under general anesthesia. After a reference frame was attached to the handle of the occlusal splint fixed with the patient's mandibular teeth, point-based registration was performed. The accuracy of the navigation system was verified, and then, the mouth was opened. A gingival incision was made, and mandibular ramus was exposed after the elevation of the mucoperiosteal flap. The navigation probe was put on the mandible, and the tip position of the probe was viewed continuously on the screen of the navigation system. Sectioning of the border between natural cortical bone of the mandibular ramus and reactive proliferative bone was performed using a calibrated ultrasonic bone device (SONOPET®, Stryker, Kalamazoo, MI) under navigation guidance (Fig. 6). After the removal of the reactive proliferative bone, the right mandibular third molar was removed from the lateral side of the mandibular ramus. To improve trismus, coronoidectomy was also performed. The wound was irrigated and closed. Intraoperative and postoperative antibiotics (ABPC: 2 g/day) were administrated intravenously for 2 weeks. After the discharge, AMPC (750 mg/day) was administrated intraorally for 7.5 months. Trismus was gradually improved, and the maximum mouth opening was finally 44 mm. Postoperative panoramic radiograph and CT showed the right mandibular angle resorption

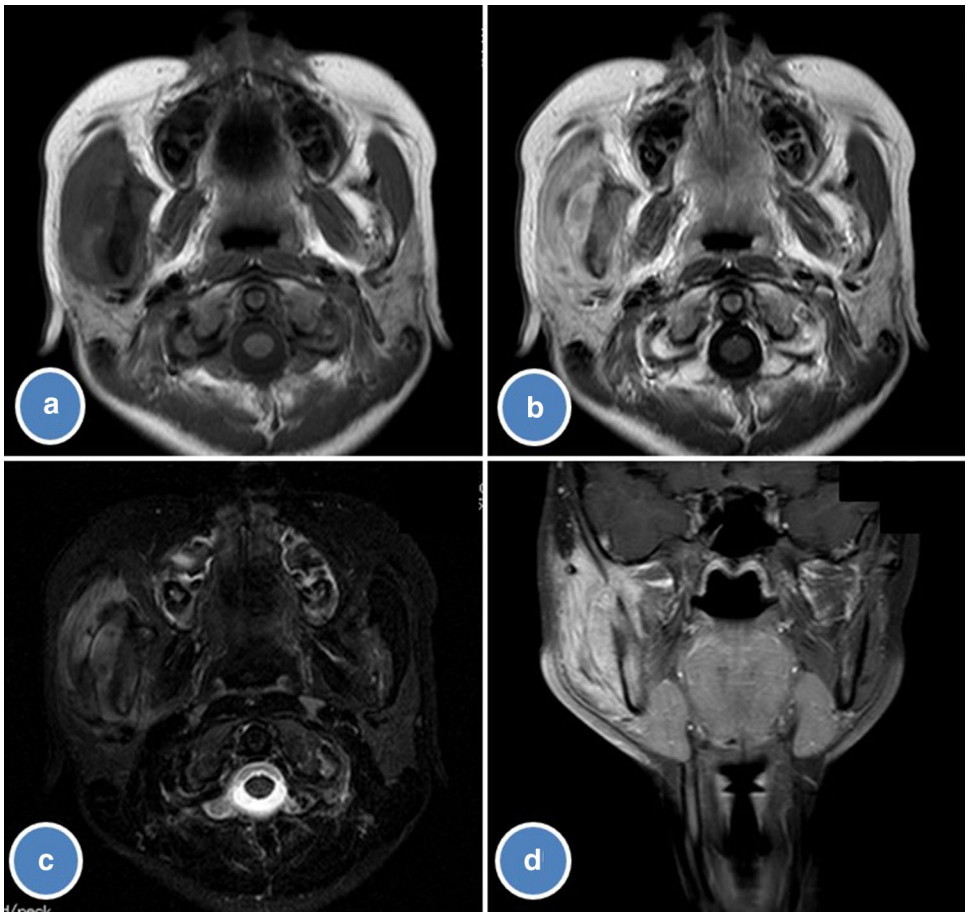

Fig. 4 MRI shows that the mandibular ramus is a low-signal intensity and the reactive bone on the ramus is signal intensity similar to muscle on T1-weighted images (**a**). Gadolinium-enhanced T1-weighted images (**b**) and short TI inversion recovery (STIR) images (**c**) reveal heterogeneous, intermediate-to-high-signal intensities of the right masseter muscle and the reactive bone. Fat-suppressed gadolinium-enhanced T1-weighted images (**d**) shows heterogeneous high-signal intensity of the right masseter muscle and the reactive bone

and coronoid process regeneration (Fig. 7). There was no recurrence of mandibular osteomyelitis 7 years after surgery (Fig. 8).

Discussion

Actinomyces that can cause chronic infections such as trauma, periodontitis, nonvital teeth, and extraction sites have been isolated from saliva, salivary calculi, carious cavity, bone sequestrum, and tonsillar crypts [4, 9, 10]. The mandible is the site of predilection of actinomycotic infection, and the posterior mandible is more frequently involved than the anterior mandible [3]. Actinomycotic lesions in the jaw, such as radicular cyst, dentigerous cyst, odontogenic keratocyst, and osteomyelitis, have been reported by many authors [2, 4, 5, 9–24]. Generally, radicular cyst or osteomyelitis can be caused by infection from peripheral sites [5, 13, 25], and can be seen to contain *Actinomyces* druses on pathological examination [2, 11, 24]. In the present case, the cause of infection was not clear, but there were *Actinomyces* druses in biopsy specimens.

Because the diagnosis of actinomycosis is difficult to mimic neoplasms and granulomatous disease, bone lesion biopsy or fine-needle aspiration is important for the diagnosis [13, 19–21]. Therefore, Sasaki et al. [23] investigated the characteristic imaging findings of actinomycotic mandibular osteomyelitis. The presence of intralesional gas, osteolytic changes with extensive inflammatory changes in the surrounding soft tissue extending to the skin surface, fistula formation, and absence of lymphadenopathy were frequently seen [23]. Furthermore, periosteal reaction and inflammation of the masseter and pterygoid muscles also may be showed. The radiological characteristics of actinomycotic mandibular osteomyelitis help narrow the differential diagnosis. Although the present case had no intralesional gas, fistula formation, lymphadenopathy, and inflammation extending to the skin surface and pterygoid muscles, there was the proliferative bone formation of the mandibular ramus, osteolytic change of the condyle, and inflammation of the masseter muscle. To our knowledge, there was only one case with

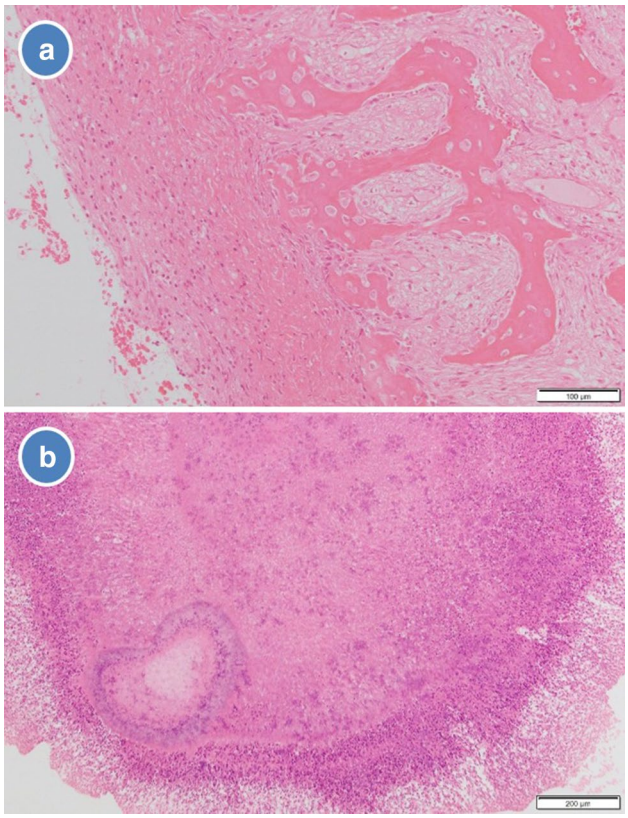

Fig. 5 Pathology (hematoxylin and eosin staining). Dense fibrous stroma in the intertrabecular space (**a**) and *Actinomyces* druse (**b**)

actinomycotic mandibular osteomyelitis with proliferative bone formation in English language literatures, excluding the present case [19].

Antibiotic treatment with penicillin and/or its derivatives is the basic form of therapy against actinomycosis. Although routine antibiotic administration for several to 12 months has been recommended [1, 3, 13, 18, 24], the modern approach to treatment is more individualized, and the exact antibiotic regimen and duration of treatment depend on the infection site, severity of the disease, and the patient's general condition and response to treatment [1, 14]. However, the treatment of actinomycosis in the oral and maxillofacial region is focused on surgical debridement including the area of the source of the infection to reduce the numbers of organisms and change the anaerobic environment that is vital for the survival of these organisms [26]. According to a review by Wong et al. [1], orocervicofacial disease has been cured after short courses of 2–6 weeks of antibiotics (oral and intravenous) combined with surgical drainage. In intraoral actinomycosis, Stenhouse et al. [9] reported that prolonged antibiotic therapy was not required when the infected material was surgically removed from the site. Similarly, Hirshberg et al. [11] stated that the outcome of patients with periapical actinomycosis was better and curettage of the lesion combined with a short course of antibiotics was sufficient to induce healing without complications in most cases. Fergus and Savord [4] reported that a patient who received antibiotics had recurring symptoms until the lesion was surgically removed. In a single institution study of 15 cases with actinomycotic osteomyelitis of the mandible [19], 20% of all patients who underwent surgery and antibiotic therapy for 3 weeks–2 months had recurrences. In a review of pediatric patients with actinomycotic mandibular osteomyelitis, all 14 cases required at least one debridement, with four of the cases requiring multiple debridement [22]. Therefore, a 3-month course of an orally administered antibiotic was recommended after the debridement [22]. These reports suggest that antibiotic treatment alone is not enough if *Actinomyces* are not eliminated and surgical treatment with or without antibiotics should be performed after early diagnosis [2, 12]. In the present case with postoperative antibiotic administration for 8 months as well as the removal of the third molar and reactive proliferative bone, there was no recurrence 7 years after surgery.

Recently, image-guided surgery with navigation systems has been applied in the maxillofacial region, especially midface. Navigation systems enable surgeons to operate safely by avoiding accidental injury of anatomical important structures, because navigation surgery allows that surgical instruments are accurately tracked three-dimensionally and targeted to a preplanned location in the surgical field. Although the mandible has been seldom included as operating fields with the navigation system for its mobility [27], we used a resin occlusal splint with titanium markers and handle to fix reference a frame for intraoral mandibular navigation surgery. The navigational guidance could allow accurate sectioning of the border between natural cortical bone of the mandibular ramus and reactive proliferative bone by tracking of the tip of the calibrated ultrasonic bone device. Belli et al. [28] also performed a navigation-guided endoscopic intraoral approach for remodeling of the mandibular condyle caused by chronic osteomyelitis with proliferative periostitis. The intraoral approach to the ramus condyle unit under navigational guidance is a minimally invasive method without visible scar and risk of facial nerve injury.

Spontaneous bone regeneration following segmental mandibulectomy or hemimandibulectomy is rare [29–33], and the incidence is reported as 2.0% (13 of 636 cases) [34]. As factors affecting bone formation, there are intact periosteum, infection, postoperative stabilization of the remaining mandibular stumps, soft-tissue protection of the

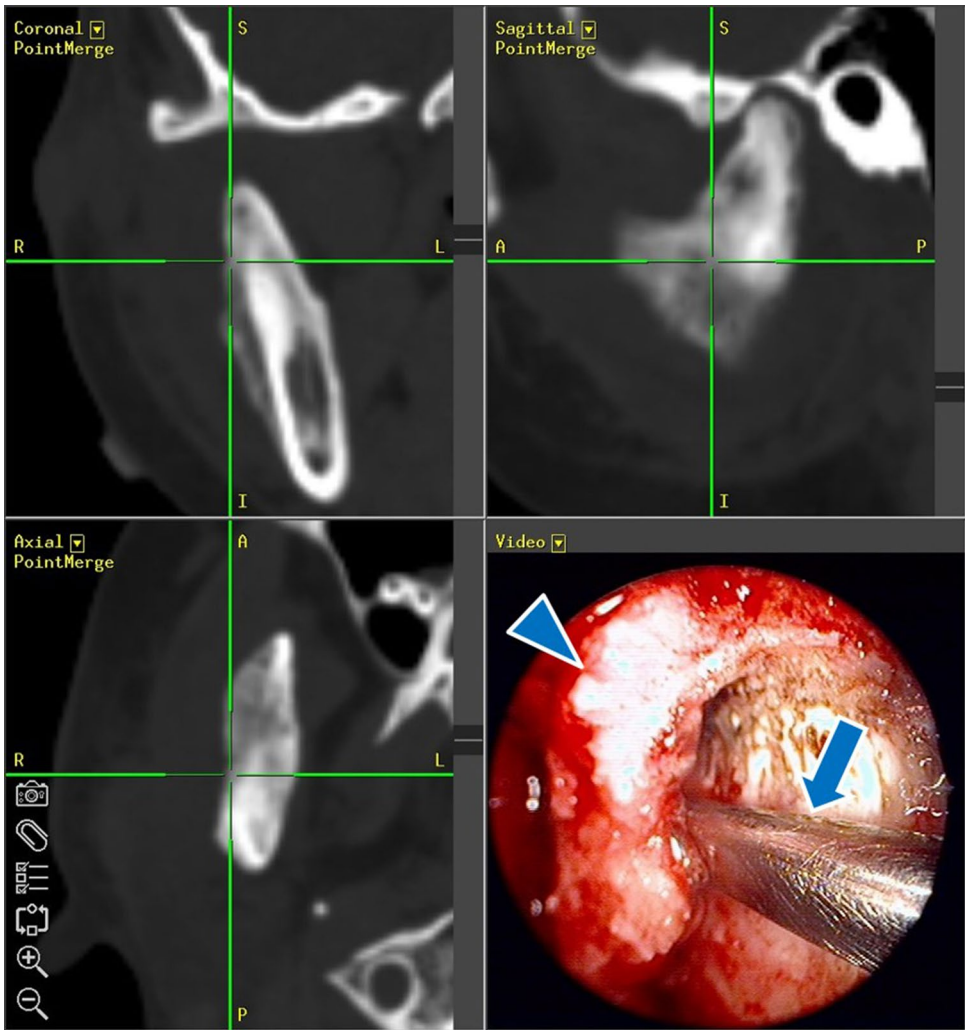

Fig. 6 Navigation monitor displaying sectioning of the border between natural cortical bone of the mandibular ramus and reactive proliferative bone (arrow head) with a calibrated ultrasonic bone device (arrow) under navigational guidance. The center of the crosshair indicates the tip of the ultrasonic bone device

bony gap, young age, genetics, among others [29–31, 34, 35]. The mandibular defect size in patients with spontaneous bone regeneration ranged from 4.7–15.3 cm, with a mean of 10.4 ± 1.8 cm [34]. Spontaneous bone formation was first evident at 2–3 months (range, 2 weeks–2 years) after segmental mandibulectomy or hemimandibulectomy [31, 34, 36–38]. The main site of bone regeneration was mandibular body, and the symphysis and parasymphysis regions were less frequently involved [31, 34]. Complete mandibular regeneration with sufficient height after segmental mandibulectomy or regeneration of well-shaped hemimandible after hemimandibulectomy is rarely reported [32, 33, 35–37, 39]. In the investigation reported by Anyanechi et al. [34], 76.9% of patients had entire mandibular regeneration (complete span) with insufficient height, whereas the remains had an incomplete bone generation. Although the age of patients with spontaneous bone regeneration is reported as 4–58 years [32–34, 36, 38], bone regeneration is promoted in children for the high cellular activity and availability of abundant mesenchymal cells to form osteogenic tissue. The younger the patients, the earlier the spontaneous bone regeneration occurred in the mandibular defect [34]. In contrast, partial preservation of the periosteum during mandibular resection plays an important role in promoting spontaneous bone regeneration in older patients comparing to the younger patients [31]. In young patients, infection, such as osteomyelitis, condensing osteitis, and proliferative periostitis, activates the osteoblasts of the periosteum [30]. Because the present case (14-year-old girl) had actinomycotic osteomyelitis with proliferative periostitis of the mandibular ramus and the periosteum was preserved during coronoidectomy, it was considered that the coronoid process of normal size was regenerated completely. Although spontaneous bone regeneration after coronoidectomy for young patients with coronoid hyperplasia was rarely reported [40, 41], generated coronoid process was normal or previous size.

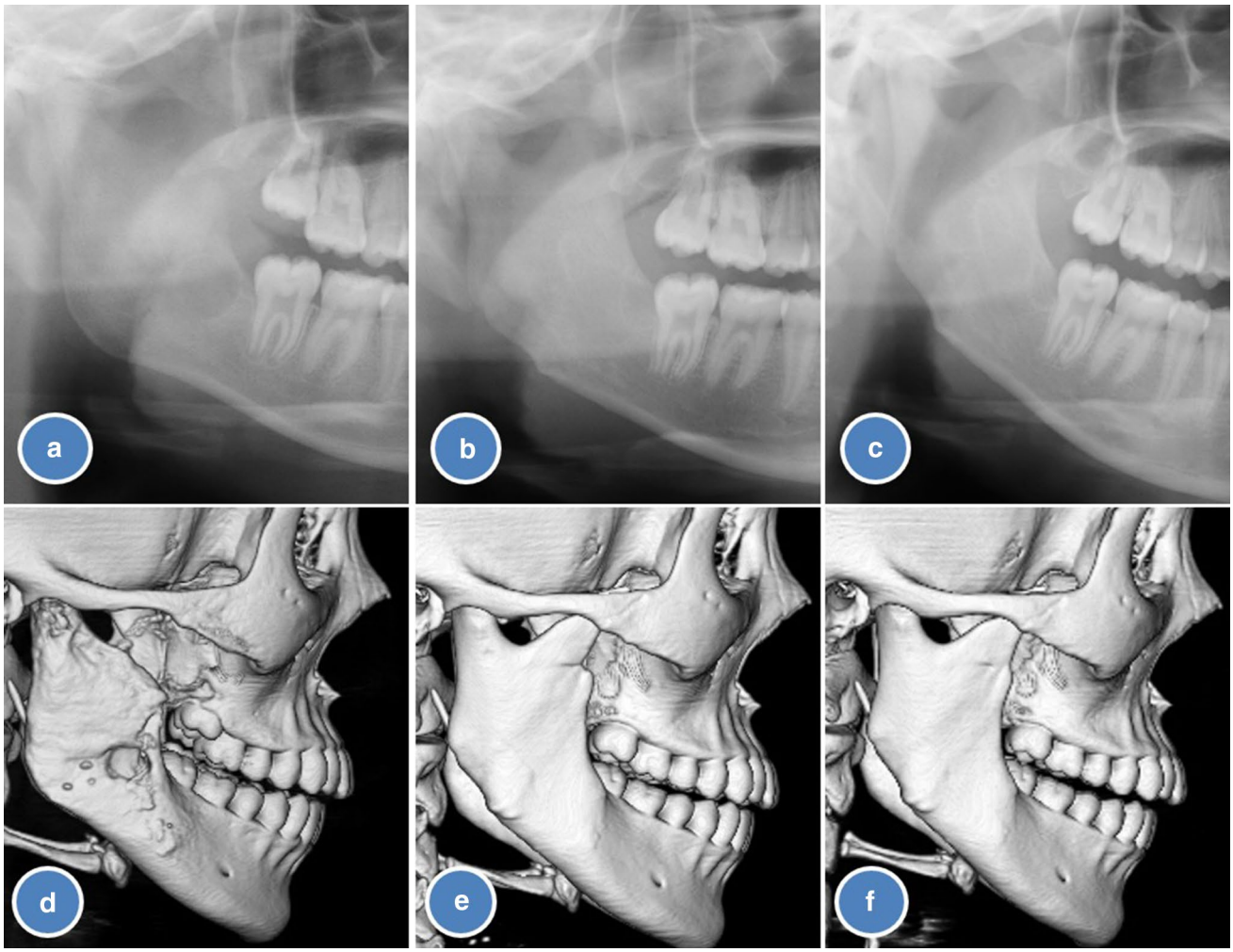

Fig. 7 Postoperative panoramic radiograph and CT shows the right mandibular angle resorption and coronoid process regeneration. **a** Panoramic radiograph 10 days after surgery, **b** Panoramic radiograph 1 year after surgery, **c** Panoramic radiograph 2 years after surgery, **d** CT 2 weeks after surgery, **e** CT 16 months after surgery, and **f** CT: 34 months after surgery

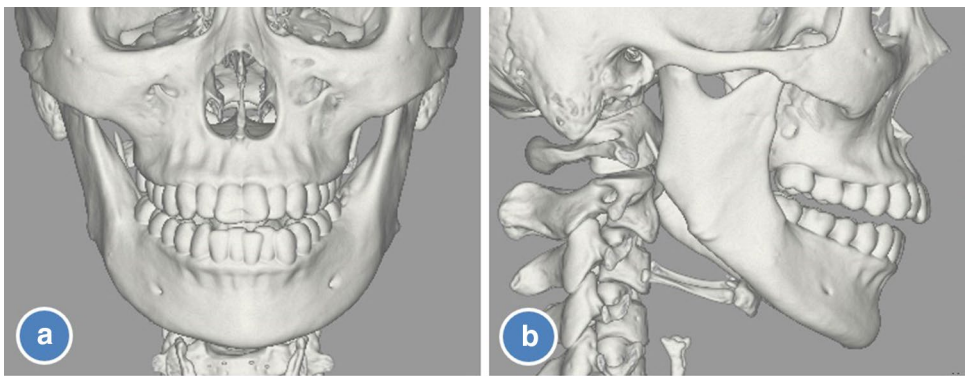

Fig. 8 3D-CT 7 years after surgery. **a** Frontal image. **b** Lateral image

Compliance with ethical standards

Conflict of interest Toshinori Iwai, Nobuhide Ohashi, Satomi Sugiyama, Hiroaki Kitajima, Makoto Hirota, Shoji Yamanaka, and Kenji Mitsudo declare that they have no conflict of interest.

Human rights statement All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Informed consent Informed consent was obtained from the patient for being included in this study.

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