J Oral Maxillofac Surg
64:981-984, 2006

Spontaneous Regeneration of Whole Mandible After Total Mandibulectomy in a Sickle Cell Patient

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Resection of the mandible and immediate reconstruction with autogenous bone graft are widely used in the treatment of ameloblastoma involving a large section of the mandible. The purpose of reconstruction is mainly to rehabilitate the patient esthetically by improving the contour of the mandible, thereby minimizing facial deformity from the defect. The patient is rehabilitated functionally and the occlusal disturbance is minimized.

Primary reconstruction by bone grafting is usually advocated at the time of surgery for various reasons. The access to the surgical site is optimal because there is no fibrosis of the graft bed. However, local facilities for surgery, infection, and patients' general condition may not often permit this. Extensive bone regeneration that reconstitutes 50%[1,2] or greater than 50% of the mandible[3-6] after injury involving a segment of mandible have been reported previously. There is, however, no reported case in which a whole mandible regenerates with condyles. This study presents a rare case of spontaneous regeneration of a whole mandible with the condyles in a 13-year-old Nigerian patient with sickle cell disease who had total mandibulectomy for an extensive case of ameloblastoma.

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0278-2391/06/6406-0020$32.00/0
doi:10.1016/j.joms.2006.02.008

Report of a Case

A 10-year-old boy reported to Oral and Maxillofacial Surgery Clinic on April 29, 1987, with a 3-week history of painless swelling of the anterior region of the mandible. The boy was a diagnosed sickle cell patient who had been attending a sickle cell clinic. Clinical oral examination showed a minimal labio-lingual expansion of the anterior region of mandible, extending from right lateral incisor to left premolar region. The lingual expansion was more pronounced. The swelling gave an eggshell cracking sensation on palpation. There was right submandibular lymphadenopathy. Intraorally, the overlying mucosa was normal whereas the lower incisors and the left canine teeth were grossly mobile. Apart from slight jaundice of the sclera, the patient did not show any other clinical manifestation of sickle cell disease. Radiographic examination of the mandible showed radiolucency extending from right canine to left premolar region. A clinical diagnosis of cystic lesion was made.

On May 18, 1987, the patient had a sickle cell crisis and was admitted under the medical unit for management. During this period, the patient complained of occasional slight pus discharge from the labial mucosa. The patient was placed on a course of Ampiclox capsules (SmithKline Beecham, Brentford, UK) (500 mg every 6 hours) for 1 week. When the medical condition of the patient was stable, an incisional biopsy was done under nasotracheal anesthesia. The pathology report came out inconclusive but suggestive of an infected cyst or ameloblastoma.

The patient defaulted from the clinic for about 3 years and reappeared in March 1990. This time, the labial expansion had increased only slightly but the radiographic examination showed an extensive radiolucent region extending from lower right second molar to the lower left second molar (Fig 1). A repeat biopsy was done on April 2, 1990, and the pathology report confirmed a cystic ameloblastoma.

The patient was admitted on May 28, 1990 for segmental resection of the mandible under nasotracheal anesthesia, after being certified fit by the medical team. At operation, on June 1, 1990, the tumor was found to have infiltrated both condyles and coronoid processes with minimal buccal expansion and no periosteal involvement. Total mandibulectomy with bilateral disarticula-

FIGURE 1. Preoperative radiograph showing radiolucency from lower right second molar to lower left second molar with bone loss around the apices of anterior teeth.

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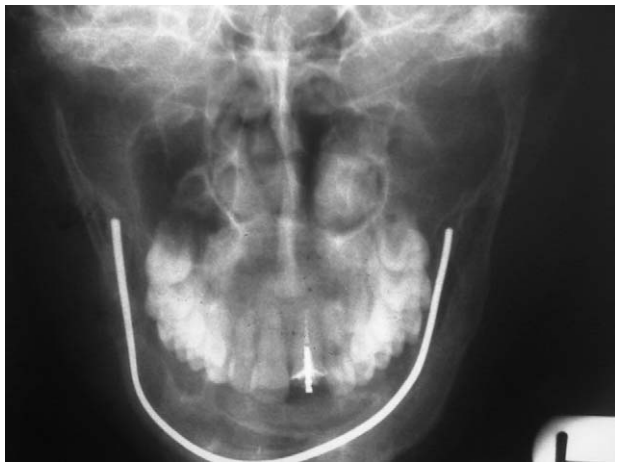

FIGURE 3. Posteroanterior radiograph of the jaws 1 year after surgery demonstrating regeneration of the entire mandible including condyles.

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tion was therefore carried out with preservation of the periosteum along the full length of the mandible. The surgical approach was intraoral. An arch bar bent to simulate the curvature of the mandible was used to preserve the surgical bed, improve the facial contour and provide support for the tongue by suturing the muscles and mucosa of the floor of the mouth over the arch bar to the lip mucosa allowing subsequent fibrosis to stabilize the arch bar. The patient was transfused with 2 pints of blood intraoperatively. The postoperative recovery was uneventful and the patient was discharged home on June 18, 1990 to be followed up on an out-patient basis.

A postoperative radiograph taken on August 13, 1990, (about 2.5 months postoperatively) showed some bone regeneration around the modified arch bar (Fig 2). Further radiographs taken on May 27, 1991 (almost 1 year later) showed complete regeneration of the mandible with well-shaped condyles (Fig 3). There was no evidence of recurrence, and the healing was complete. The facial contour was very satisfactory, and patient was lost to follow-up again only to reappear in 2002 with a request for replacement of the teeth when a fresh radiograph was taken (Fig 4).

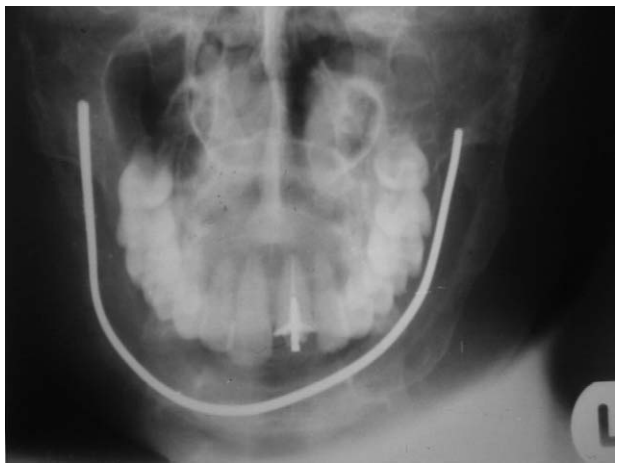

FIGURE 2. Posteroanterior radiograph of the jaws 2.5 months after surgery demonstrating regeneration of bone.

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Discussion

New bone formation can take place through the process of osteoinduction, osteoconduction, and osteogenesis.[7] Osteogenesis is the formation of new bone from osteoprogenitor cells in a wound. The osteoprogenitor cells that differentiate into osteoblasts are found within the bone marrow, endosteum, and the inner cambium layer of the periosteum.[8] The role of the periosteum as an important structure in fracture healing and bone grafting is recognized,[5,9] and it is important, therefore, to preserve it. It is also believed that the periosteum plays a role in spontaneous bone regeneration.[1-7] Even irradiated periosteum still has some osteogenic potential although to a lesser degree than non-irradiated tissue.[5] Rugerrio and Donoff[5] reported a case of spontaneous regeneration of the mandible after irradiation.

The case described in this study supports the important role of the periosteum in spontaneous regeneration. Total mandibulectomy with bilateral disarticulation was carried out in this case without any bone graft. The possibility of contributory osteoprogenitor cells from other sources (ie, the endosteum or bone matrix) is ruled out because there was no portion of the mandible left and bone grafting was not done. An arch bar was contoured and placed in the surgical bed to maintain the space and improve the patient's appearance. The only possi-

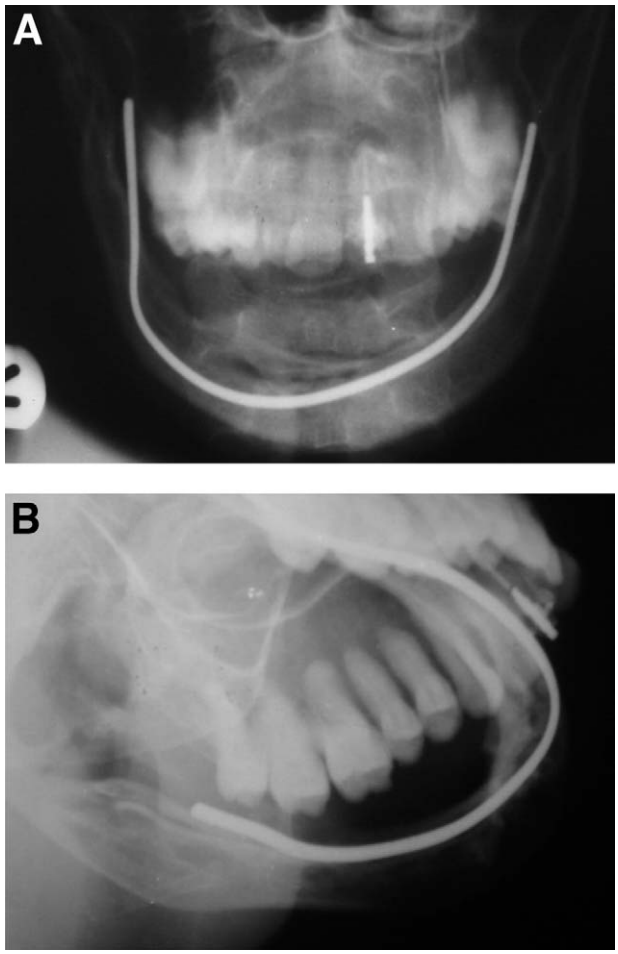

FIGURE 4. Radiograph of the jaws. *A*, posteroanterior view. *B*, oblique view showing the regenerated mandible 10 years postoperatively.

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ble source of new bone in this case is likely to be the periosteum.

Spontaneous regeneration of a large portion of the mandible had been reported previously.[1-5] In these reports, regeneration was subsequent to hemimandibulectomy or subtotal mandibulectomy, but not total mandibulectomy as reported in this case. Spontaneous regeneration of a lost portion of bone is not limited to the jaws. It has also been reported in long bones after injuries.[6] Spontaneous regeneration of the whole mandible with well-shaped condyles occurred in a sickle-cell patient without the need for bone graft. The striking thing about this case is that regeneration was noticed 2.5 months after the operation and by 1 year after there was regeneration of the entire mandible with well-shaped condyles. The complete regeneration of the entire mandible with both condyles in such a short length of time has not been reported previously in literature. There is, however, a report of spontaneous regeneration of half of the mandible with well-shaped condyle.[2]

The exact mechanism of induction of periosteal bone regeneration is not fully understood but several factors have been suggested to influence this. These include age of the patient,[2-6] infection,[3,4] damage to the periosteum,[2,4] increased tension in the bone, and genetic behavior.[4]

Cases of spontaneous regeneration of the mandible reported in the literature are in young individuals with the age ranging from 5 to 11 years[1-5,7] with the exception of a reported case in a 27-year-old patient.[5] The cases reported from Nigeria previously[3,4] are in 12- and 15-year-old patients. Spontaneous regeneration of the entire lateral malleolus including the epiphysis, physis, and metaphysis after injury has also been reported in a 3-year-old patient.[6] The case described in this study occurred in a 13-year-old patient. This suggests that age may play an important factor in spontaneous regeneration due to high cellular activity and availability of abundant mesenchymal cells of children to form osteogenic tissue.[6] The cause of spontaneous regeneration in some individuals and not in others of the same age is still not fully understood.

Infection is believed to be a stimulating factor in periosteal bone regeneration.[3,4] It is believed to activate the osteoblasts from the periosteum so that when it is left intact, bone regeneration can occur. Presence of infection either preoperatively,[3] intraoperatively,[3] or postoperatively[5] is associated with reported cases of spontaneous regeneration of the mandible. In our case, the lesion was infected 3 years before the surgery was carried out. It is therefore debatable whether this infection could have been a stimulant for the periosteal bone regeneration that occurred 3 years later. One is not sure, however, whether there had been repeated infection of the lesion during the patient's absence from the clinic.

Stress[4] is another factor that has been implicated in spontaneous bone regeneration. In cases where spontaneous regeneration was subsequent to removal of a certain portion of the mandible, immobilization of the remaining segment has been suggested as promoting osteogenesis.[5,6] There was no immobilization in this case because total mandibulectomy with bilateral disarticulation was carried out. However, an arch bar was placed in the surgical bed to improve the aesthetics of the patient.

Because the exact factors that enable the periosteum to regenerate are not fully understood, it is suggested that the periosteum should always be preserved when carrying out mandibulectomy for benign lesions that have not perforated the lingual and buccal cortical plates.

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J Oral Maxillofac Surg
64:984-988, 2006

Imaging (Computed Tomography, Magnetic Resonance Imaging, Ultrasound, Sialography) in a Case of Recurrent Parotitis in Children

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Recurrent parotitis in children (RPC), or juvenile parotitis with sialectasis, is an infrequently occurring salivary gland inflammatory disease observed in children, with its highest incidence between 3 and 6 years of age.[1] The condition involves males slightly more often than females[2-8] and has a marked tendency to regress with puberty.[2,6,9-11] However, clinical remission may not occur until early adult life,[4,12] and continuation throughout adult life is always a possibility.[1,10]

Characteristically, both parotid salivary glands are involved even if the subjective symptomatology is initially apparent on only 1 side. Subsequent episodes can alternately implicate the opposite side, and at times the exacerbation can simultaneously involve both parotids. The parotid swellings are sudden in onset, unrelated to eating, and may be accompanied by pain and fever. Recurrences are common, with flareups tending to occur 1 to 5 times each year.[3,4] Each episode lasts approximately 3 to 7 days,[3] and tends to subside with or without treatment. Neither the submandibular or sublingual salivary glands are involved in the disease process.

During periods of remission, salivary production is normal with no xerostomia evident.[5,13] However, during the active phase, salivary volume may be diminished.[14] More significantly, during an exacerbation milking of the involved gland will produce a saliva that contains flocculations. These flocculations represent the presence of elevated levels of salivary proteins, mucus, inflammatory cells, and even some pus when infection develops as a secondary manifestation.[2] The infection is a consequence of an ascending ductal invasion by bacteria, facilitated by salivary stagnation, and failure of adequate ductal lavage. Clinical recovery occurs between periods of parotid swelling and a normal clear salivary flow can be observed.

Histologically, RPC is characterized by dilatation of the interlobular ducts and a periductal lymphocytic infiltration.[8,15,16] The etiologic mechanism that initiates this histologic process is not understood, and an accepted origin remains an enigma.

The diagnostic hallmark of RPC has been the sialographic demonstration of sialectasis coupled with the

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0278-2391/06/6406-0021$32.00/0
doi:10.1016/j.joms.2005.11.057