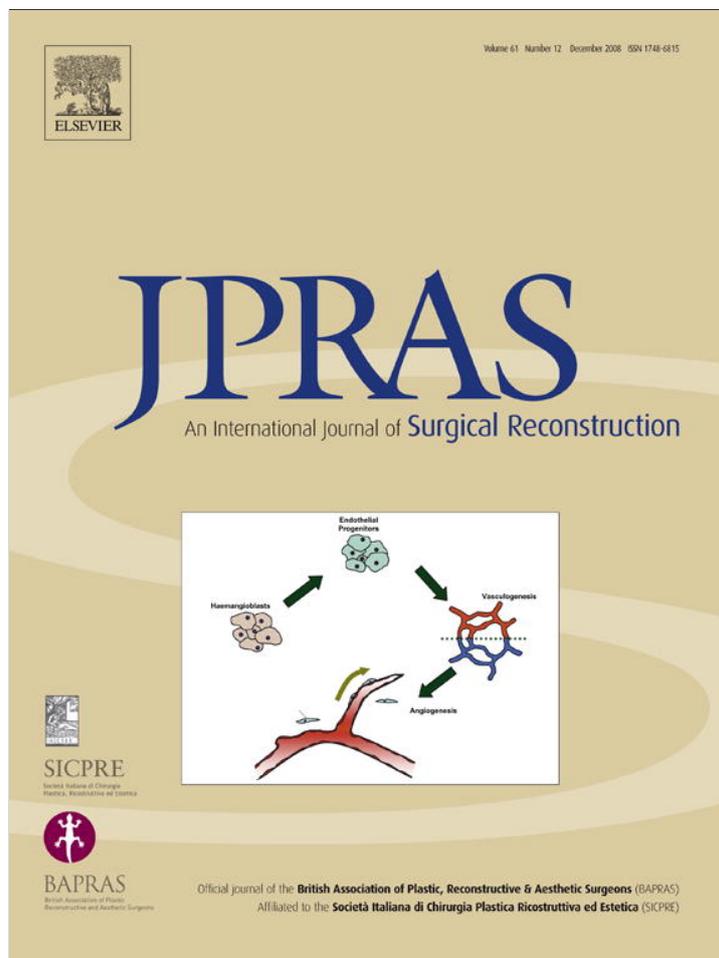


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CASE REPORT

Reconstruction of a huge oral maxillofacial defect caused by necrotic fasciitis secondary to leukaemia

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Summary An 8-year-old boy who was diagnosed with 'acute non-lymphoblastic leukaemia (M2)', contracted secondary oral maxillofacial necrotic fasciitis. The wound was cleaned with 3% hypertonic saline, and then covered with iodoform gauze every day for about 3 weeks before and after necrotic tissue debridement. The local infection was controlled, and plenty of new healthy granular tissue had grown. The patient was left with a huge defect including the mouth floor, submental area, submandibular area and right cheek when the necrotic tissue was removed. Reconstruction of the defect was very difficult because of the patient's pre-existing leukaemia condition and severe inflammatory local condition. We successfully reconstructed the defect by using the new healthy granular tissue and a trapezius myocutaneous flap. During 9 years of follow up, in order to improve quality of life, the patient underwent scar modification surgery and orthodontic treatment, and facial appearance and oral functions were deemed satisfactory.

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Leukaemia is a malignant haematological disease. Anaemia, fever and haemorrhage are the main symptoms. Most of the time, the fever is suggestive of infection.

Stomatitis, gingivitis, and so on, often appear in these patients, and can lead to ulcers, necrosis and even sapraemia. Successful treatment consists of early diagnosis, intensive systemic antibiotherapy, aggressive surgical debridement and control of the underlying disease. When the amount of necrotic tissue removed is huge, there will be a tissue defect and its reconstruction is a big challenge. We now report one such case that was followed up for 9 years.

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Case report

An 8-year-old boy was diagnosed with 'acute non-lymphoblastic leukaemia (M2)' based on marrow and immunological examination at Peking University, People's Hospital 2 years previously. He complained of fever, gingival bleeding, submandibular space infection and swelling during his 10th chemotherapy session. A tracheotomy had been performed because of dyspnoea. Effusion, canker and necrosis of the tissue appeared as the infection became more and more severe. After consultation with the oral and maxillofacial surgery department in our hospital, the patient was diagnosed with 'secondary oral maxillofacial necrotic fasciitis' (Figure 1a).

While intravenous antibiotherapy was given, the wound was also cleaned with 3% hypertonic saline, and then covered with iodoform gauze every day. One week later, the necrotic tissue was separated from the wound. A huge defect of the mouth floor, submental area, submandibular area and right cheek was left after the wide surgical debridement of the necrotic tissue. The right mandibular body and bilateral submandibular glands were completely

exposed, the tongue prolapsed and saliva effused (Figure 1b). The same treatments were continued for 1 week, and new healthy granular tissue began to grow gradually. In order to cover the exposed mandible, some small holes were drilled into it and, 1 week later, the granular tissue had grown into the holes, and covered the mandible (Figure 1c, d). At the same time, the patient's general condition was well and stable, body temperature, results of blood coagulation and immunological protein examinations were all normal. Consultation with the paediatric department of the People's Hospital and Plastic Surgery Hospital and our hospital suggested that it was the proper time for surgical reconstruction of the maxillofacial defect.

When the patient's preoperative preparation was complete, the trapezius myocutaneous flap (10 × 35 cm) was designed to reconstruct the huge defect in the oral maxillofacial area. The flap was designed as shown in Figure 2a originally, and the top end of the flap was folded in order to reconstruct the mouth floor defect but, during the surgery, we found that the soft tissue of the mouth floor and abdomen of the tongue could be sutured

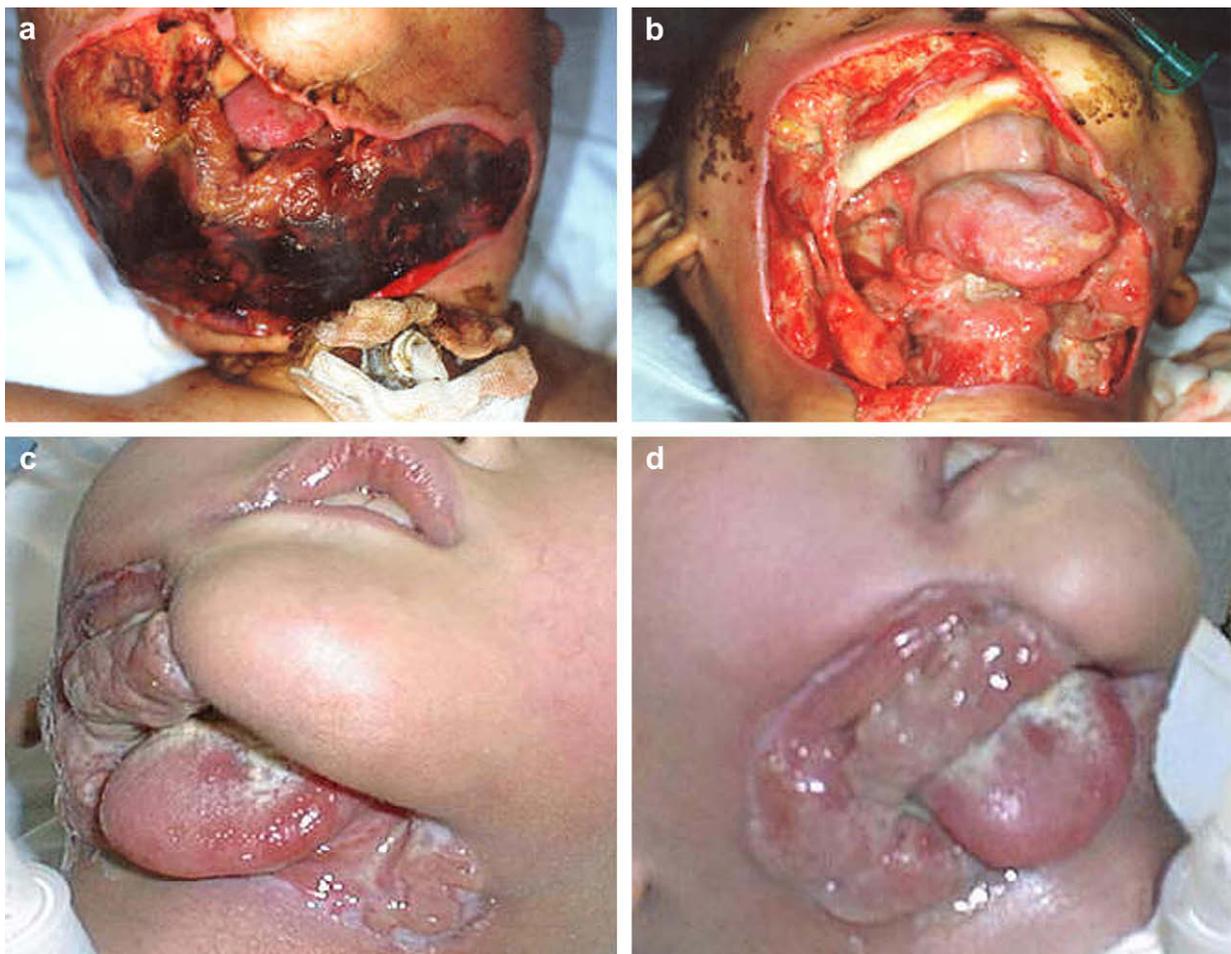


Figure 1 (a) The patient was diagnosed with secondary oral maxillofacial necrotic fasciitis. (b) A huge defect of the mouth floor, submental area, submandibular area and right cheek was left after the surgical debridement. (c, d) New healthy granular tissue began to grow gradually, and covered the mandible.

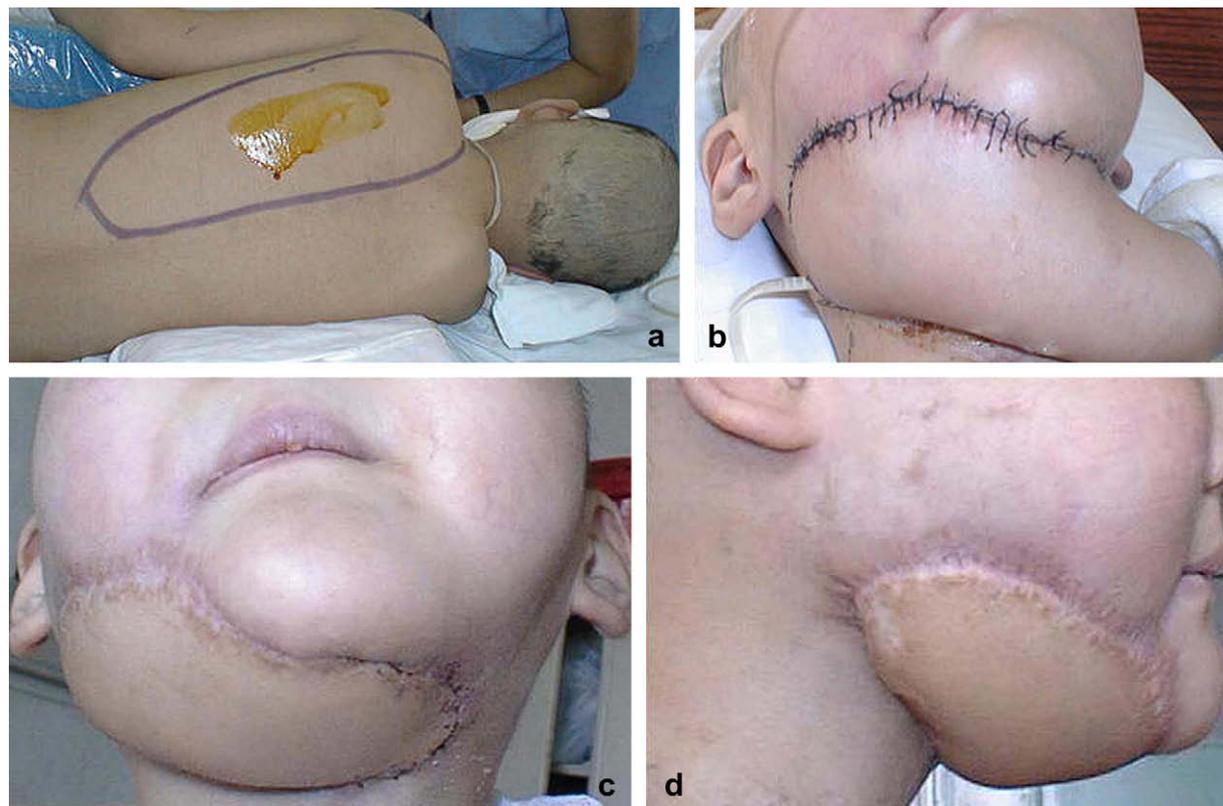


Figure 2 (a) The trapezius myocutaneous flap (10 × 35 cm) was designed to reconstruct the huge defect in the oral maxillofacial area. (b) The trapezius myocutaneous flap was just used to reconstruct the extraoral defect. (c) Twenty-five days after the surgery, the pedicle of the flap was cut off. (d) Ten days after the pedicle was cut off, the facial wound was healing well.

with the new healthy granular tissue which was on the lingual side of the mandible. So the trapezius myocutaneous flap was just used to reconstruct the extraoral defect (Figure 2b). The surgery was successful, and 25 days later, the pedicle of the flap was cut off (Figure 2c). Ten days after the pedicle was cut off, the facial wound was healing well (Figure 2d), and the dorsal wound cicatrised. The patient was discharged 1 week after removing the tracheal tube.

One year later, all of the patient's leukaemia test results were normal, the reconstruction of the facial defect was satisfactory (Figure 3a, b) and pronunciation was clear. However, the margin of the flap had a clear scar (Figure 3c), tongue movement was still limited (Figure 3d), and occlusion was very bad (Figure 4a, b). Two years later, the oral maxillofacial scar was modified by surgery, and the malocclusion was treated by an orthodontist in our hospital for 4 years.

Nine years later, leukaemia test results, aesthetic contour, occlusion, tongue movement and pronunciation were normal (Figure 4c, d), and the patient has finished his middle school education.

Discussion

Fever caused by infection is one of the main symptoms for a leukaemia patient. The infective symptom presents

atypically, and it is prone to become sapraemia. Sustained and large dosage chemotherapy accelerates the damage to normal tissue, such as skin and mucosa, and this makes it easier to acquire infections. Most deaths from leukaemia are actually caused by infections. Severely infected tissue often becomes necrotic, which needs to be totally removed by surgery, and the resulting, sometimes huge defects need to be reconstructed.¹⁻⁴ Because of the poor general condition of leukaemia patients, suffering from conditions such as anaemia, cacotrophia and so on, not only infection control but also tissue regeneration and reconstruction are difficult.

In this case, the patient had caught a severe infection during his 10th chemotherapy session, and the infection developed into oral maxillofacial necrotic fasciitis. The necrotic tissue could not be removed because of bleeding and sapraemia until it had been separated from the normal tissue completely. What is more, plenty of effusion and saliva on the wound surface made the infection hard to control. So at this stage, the main treatments were systemic infection control and local effusion decrease. Hypertonic saline is often used to dress acute inflammatory wounds in order to decrease the local tissue oedema and the amount of effusion, and at the same time inhibit bacterium growth.⁵ It can be used once or twice, even three times a day depending on the condition of the wound. Iodoform gauze has been used in various wounds of the body and, although there are some side effects,⁶ it is

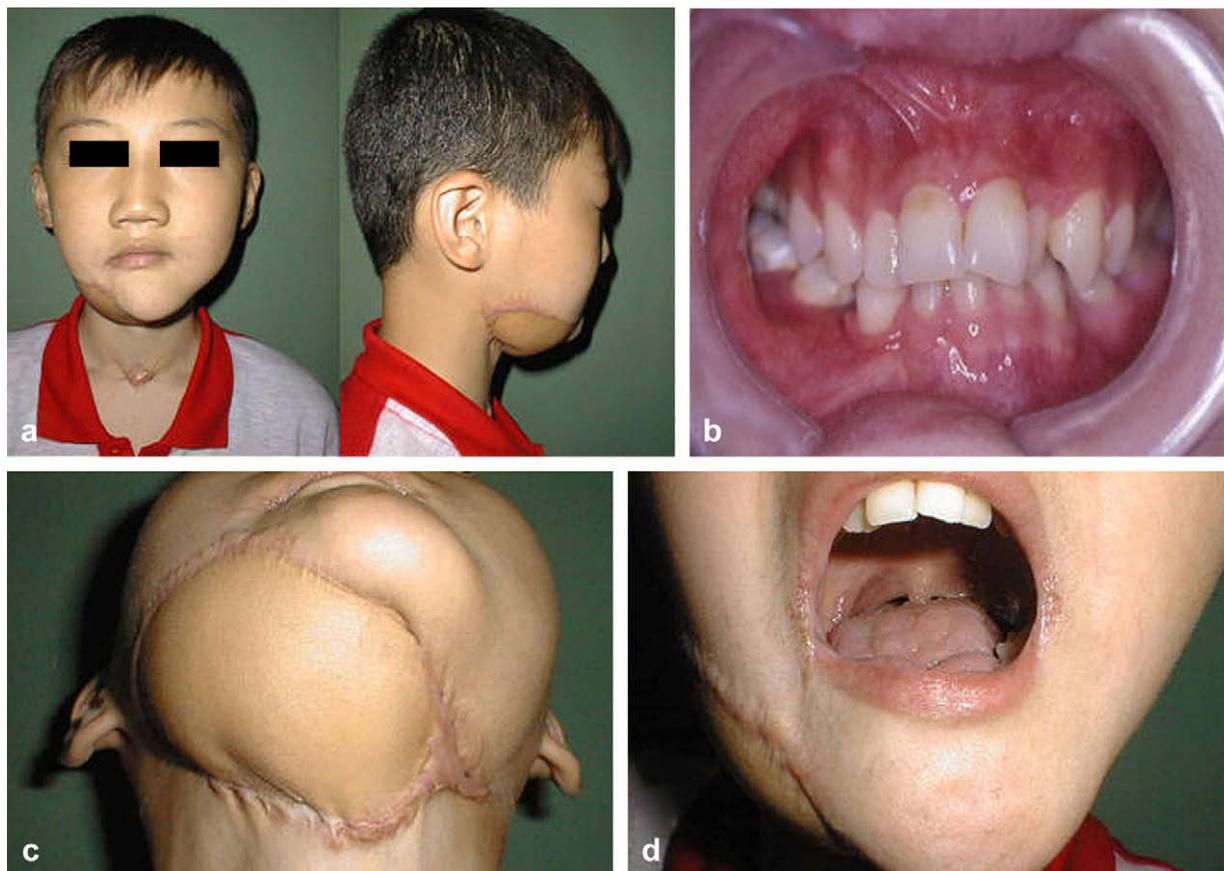


Figure 3 (a, b) One year later, the reconstruction of the oral maxillofacial defect was ideal. (c) The margin of the flap had a clear scar. (d) Tongue movement was still limited.

harmless for most human beings.⁷ Iodoform releases iodine when it comes into contact with bodily fluids, which has a powerful sterilisation effect, including bacteria, viruses, epiphytes and sporangium. Meanwhile, iodoform gauze can also absorb the effusion, staunch the wound bleeding, and keep the wound dry. All of these are very important for wound healing and new healthy granular tissue regeneration. From this case, we can see that hypertonic saline and iodoform gauze did help to control the wound infection locally, and the necrotic tissue was separated 1 week later, with new healthy granular tissue regenerated 2 weeks later.

The new healthy granular tissue played an important part in this patient's reconstructive surgery. After the necrotic tissue was removed, a huge defect was left, including the right mandibular surface, and it is very important to protect this bone from marginal osteomyelitis. Because of the favourable growth of the new healthy granular tissue, we decided to cover the mandible with it. In order to help the granular tissue to cover the smooth surface of the mandible, some small holes were drilled into the mandibular marrow. There has been no research on this method, but it is well known that the soft tissue will grow into the holes first, compared with bone formation, so we felt this soft tissue would provide a good scaffold of soft tissue for more granular tissue growth. The result was very satisfying, and the whole mandible was covered, and

protected from marginal osteomyelitis. During the reconstructive surgery, the new healthy granular tissue also provided enough soft tissue for reconstruction of the oral side defect. It made the surgery much easier and decreased the surgical trauma.

It was vital to choose the right time for this leukemia patient's reconstructive surgery. The patient's local and general condition should be both well and stable.

The trapezius myocutaneous flap has been widely used to reconstruct huge defects and to release tight scars of the head and neck.^{8–10} Although this flap has some disadvantages, i.e. an obvious scar on the back and another operation to cut the pedicle and remove excessive tissue from the face, we decided to use it because the defect was huge, including oral mucosa and facial skin, and the tissue and vessels of this leukaemia patient were abnormal after his 10th chemotherapy session.

The goals of reconstruction include not only aesthetic contour, but also restoration of oral function, e.g. mastication and speech. These are very important, especially for an 8-year-old boy. His oral-facial defect has been reconstructed successfully and, during the 9 years of follow up, he also underwent other treatments, scar modifying surgery and orthodontic treatment, to improve his aesthetic contour and oral functions, and to give him the best possible quality of life.

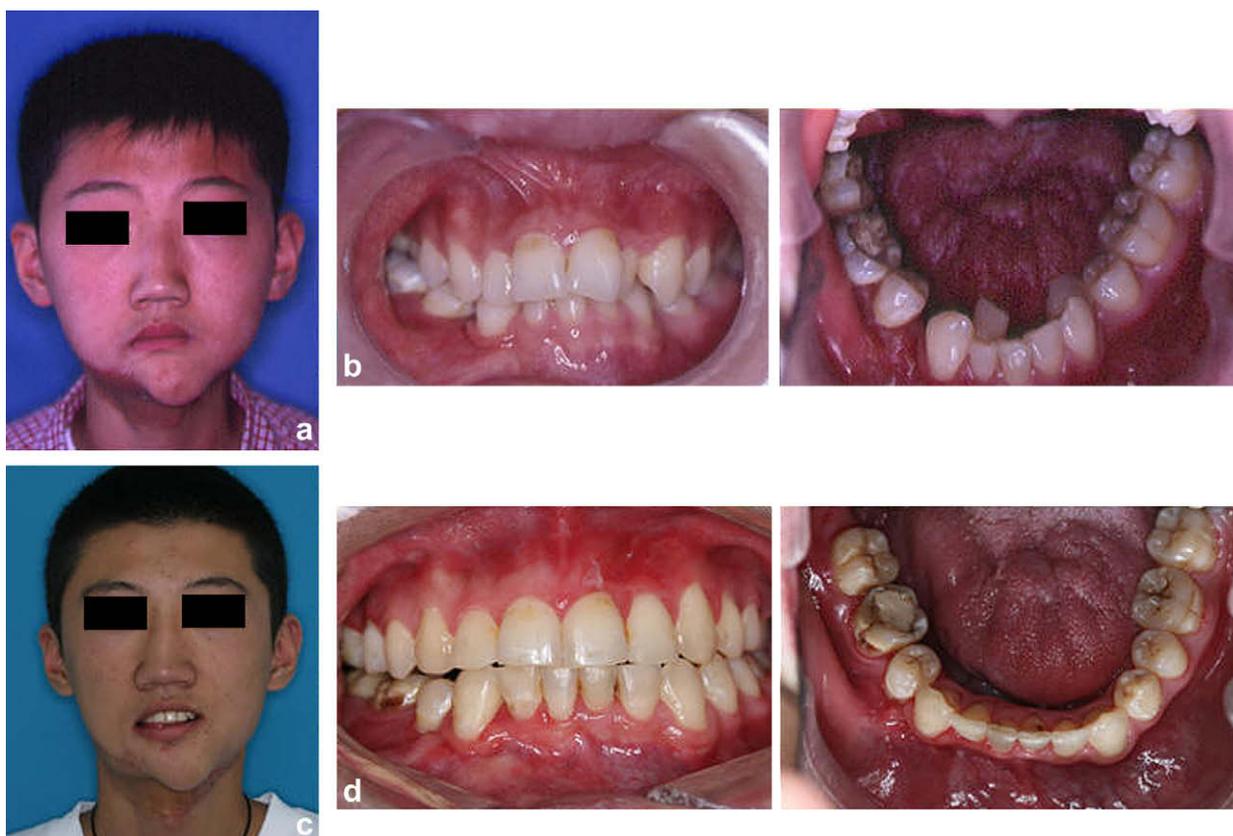


Figure 4 (a, b) Before orthodontic treatment, the patient's occlusion was very poor. When he was 10 years old, the malocclusion was treated by the orthodontist in our hospital for 4 years. (c, d) Nine years later, when he was 17 years old, the aesthetic contour, occlusion, tongue movement and pronunciation were normal.

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