Clinical Case Report

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Surgical correction of residual facial deformity following conservative excision of a giant maxillary ossifying fibroma

A case report

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Abstract

Rationale: Ossifying fibroma (OF) is a benign fibro-osseous lesion that can develop in the oral and maxillofacial region. OF is more common in females and has a marked predilection for the mandible, occurring rarely in the maxilla. Lesions grow slowly and are usually asymptomatic until growth produces an obvious swelling, pain, paresthesia, and facial deformity. With low rates of recurrence, treatment is usually curettage or resection. Very large lesions that invade other organs and that cannot be completely removed should be excised conservatively.

Patient concerns: We present a case of a 46-year-old female with a very large fibro-osseous lesion arising from the maxilla who was more concerned about the facial appearance and requested conservative treatment for economic reasons.

Diagnoses: The pathological results based on conservative excision of the lesion confirmed the diagnosis of OF.

Interventions: We chose conservative excision via the Weber–Ferguson approach and followed up every 6 months. Facial deformity correction was performed 2 years postoperatively and right lower eyelid ectropion correction 3 years after the primary excision.

Outcomes: The ectropion deformity in the right lower eyelid improved dramatically with a better facial appearance and no obvious swelling.

Lessons: Treatment programs for OF should be individualized based on the size, growth rate, invasion, and interference with facial function and esthetics. If lesions grow slowly, we suppose that it is feasible to excise conservatively when reconstruction cannot be performed due to esthetic and functional problems. Also regular postoperative follow-up is necessary to detect recurrence, and to improve facial appearance as required.

Abbreviation: OF = ossifying fibroma.

Keywords: facial deformity, facial esthetics, giant maxillary ossifying fibroma, maxilla, ossifying fibroma

1. Introduction

Ossifying fibroma (OF) defines a group of benign osseous lesions characterized by a high number of complex and variable inorganic minerals histopathologically.[1] OF may have an odontogenic origin,[2] and is believed to derive from pluripotent cells capable of differentiating into lamellar bone, cementum, and fibrous tissue.[3]

OF has been reported in all age groups, from children to adults, and occurs most frequently in females 36 to 48 years of age.[4,5] OF manifests as an intraosseous and asymptomatic mass that grows slowly and eventually causes progressive destruction and deformation of surrounding bone.[6] OF is more likely to occur in the mandible than in the maxilla if it develops in the jaw.[5]

Diagnosing OF depends on its clinical manifestation, microscopic features, and radiological features. Microscopic examination usually reveals spherules or trabeculae of bone or cementum mixed with cellular fibrous connective tissue matrix. Radiographically, the internal structure appears as a mixture of radiolucent and radiopaque densities depending on the quantity and form of the calcified material.[7] Differential diagnoses include osteoblastoma, chronic sclerosing osteomyelitis, osteoid osteoma, Pindborg tumor, ameloblastoma of the maxillary sinus, calcifying odontogenic cyst (Gorlin cyst), osteogenesis imperfecta, odontogenic myxoma, and Paget disease.[8]

Maxillary OF is rare, with few reports in the literature. Recurrence is variable and surgical excision is the preferred treatment.[4,9] A definitive surgical protocol for OF is lacking,

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and only a small number of reports discuss recurrence and facial deformity with follow-up. We present a 46-year-old female patient with a giant maxillary OF who underwent conservative resection. Within 3 years after surgery, she had 2 operations to correct facial deformity to improve her appearance. The aim of this report was to provide a basis to develop clear surgical protocols as well as to emphasize the importance of improving facial appearance through multiple follow-ups and treatments.

2. Case report

A 44-year-old female patient presented to our department in November 2013 with a chief complaint of a giant swelling on the right side of her face that had gradually increased in size over 30 years. She experienced no related pain, paresthesia, or tenderness. On examination, the swelling in the right maxillary region measured $15 \times 15 \times 10$ cm in size and had normal skin appearance. Additional findings included apparent drooping of the upper lip with a widened philtrum, distorted appearance of the nose and external nares, disappearance of the right alar groove, exposed right palate, and partially exposed teeth on the right side. There was no associated temperature or color change and no tenderness, but the skin surface was thickened. Inside the oral cavity, the maxillary lesion occluded the right buccal and labial vestibules, and the right buccal vestibule and palate were expanded with a normal mouth opening and shifted maxillary teeth with redundant oral mucosa (Figs. 1 and 2).

Computed tomography scan and 3-dimensional reconstruction revealed a $13 \times 13 \times 9$ cm expansile lesion arising from the right maxilla with defined anterior margin and rough surface. Superiorly, the lesion was eroding the right inferior orbital wall and posterosuperiorly, the lesion had displaced the maxillary sinus, which appeared shrunken. The lesion involved the right os nasal medially and the right alveolar bone distally, and appeared as a solitary radiolucent cyst-like mass with irregular radiodensity. The cortex and medulla were not clearly separated (Figs. 3 and 4).

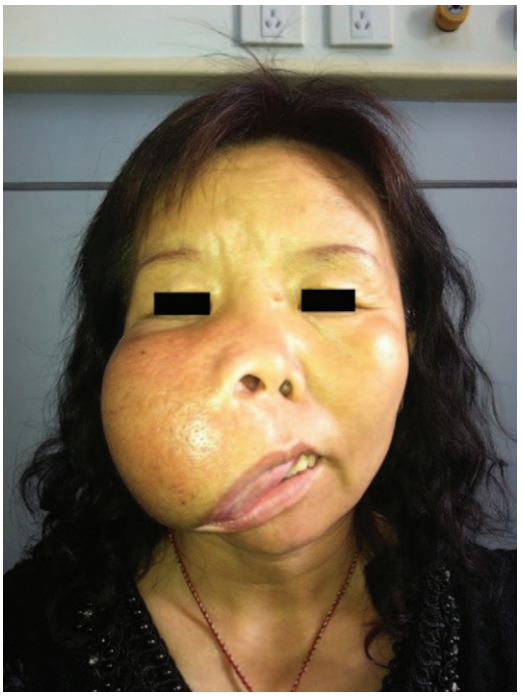

Figure 1. A patient with a giant swelling on the right side of her face that had gradually increased in size over 30 years.

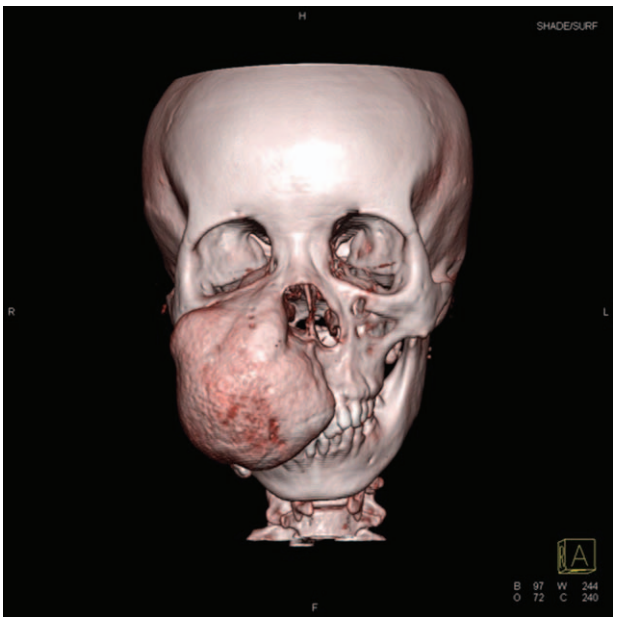

Figure 2. Frontal view of three-dimensional reconstruction.

Because of the size of the maxillary tumor, reconstruction was necessary after complete resection to maintain the function of adjacent organs that had been invaded by the tumor. However, the patient refused bone or metal grafting for financial reasons and was treated with conservative excision after exposing the tumor using the Weber–Fergusson approach. We removed as much tumor tissue as possible while maintaining functional bone tissue and excised the excess skin and oral mucosa (Fig. 5). Histopathologically, the lesion appeared as trabecular bone surrounded by osteoblasts scattered within proliferative fibrous tissue. Trabecular bone was thickened and arranged in parallel sheets with surrounding calcification, which confirmed the diagnosis of OF (Fig. 6).

Two years postoperatively, in November 2015, the patient complained of conspicuous scars and drooping of the upper lip on the right side with no visible tumor swelling (Fig. 7). To improve her facial appearance, we performed a second surgery using the same approach to excise scar tissue and partial residual tumor, correcting the droop in the right upper lip. During recovery, local infection caused by bone wax rejection leads to subcutaneous adhesion of the right lower eyelid, which finally resulted in right lower eyelid ectropion through scar contracture (Fig. 8). In February 2017, with no obvious increase in volume of the tumor, the patient underwent corrective surgery for the valgus deformity in the right lower eyelid. We separated the subcutaneous tissue to relax the constricted scar and implanted a crescent-shaped silicone prosthesis to avoid recurrence of subcutaneous adhesions and scar contracture (Fig. 9). Following this third surgery in January 2018, the ectropion deformity in the right lower eyelid improved dramatically and no obvious swelling of tumor was found (Fig. 10); however, the patient was still followed for complications and tumor recurrence regularly.

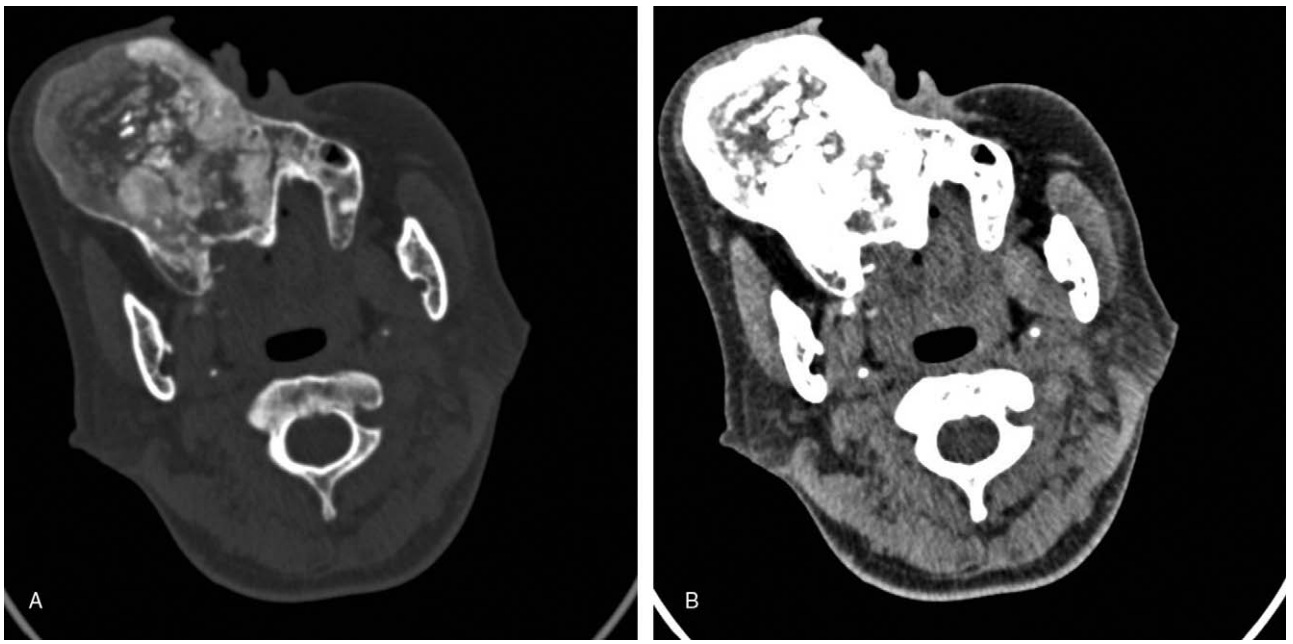

Figure 3. (A, B) Bone and soft tissue window of computed tomography axial view shows a well-defined lesion with radiolucent and radio-opaque foci.

The patient gave signed consent to publish the details of her case.

3. Discussion

OF originates from periodontal ligament cells, which develop from mesenchymal cells that form bone and cementum.[10,11] OF can develop in various forms of bone, such as cementum, woven, or lamellar bone, and appears as a widely variable solid mass, with different clinical and microscopic manifestations.[1] The etiology of OF remains unclear. Most believe that OF formation is linked simply to a disturbance of bone maturation of congenital origin,[12] and some suggest that OF arises from a stimulus provided by previous tooth extraction or periodontitis.[6] The lesions are most commonly seen in the third and fourth decades of life with female preponderance. OF occurs more often in the mandible than the maxilla. Maxillary OF is rare, with only a few reports in the literature.

In our patient, the OF was giant and measured $15 \times 15 \times 10$ cm, which is quite rare. Computed tomography scan and 3-dimensional reconstruction revealed extensive involvement of surrounding tissue, which increased the surgical difficulty. Similar recent case reports with different treatments and follow-up results are rare in the published literature. The treatment for OF is usually curettage or resection, and complete removal of the lesion as soon as possible is the approach suggested by the majority of the authors.[13–15] Titinchi and Morkel[16] reviewed 61 cases of OF from a tertiary hospital in South Africa and believed that curettage is an acceptable surgical procedure with low recurrence rates while resection should be reserved for recurrent and aggressive lesions. Currently, there is no uniform standard for the treatment of giant facial OF involving variable facial organ dysfunction. Ram et al[17] suggested that the OF lesion be excised conservatively in a case of giant maxillary OF in a 45-year-old female patient with ectropion of the right eye. However, others have suggested that

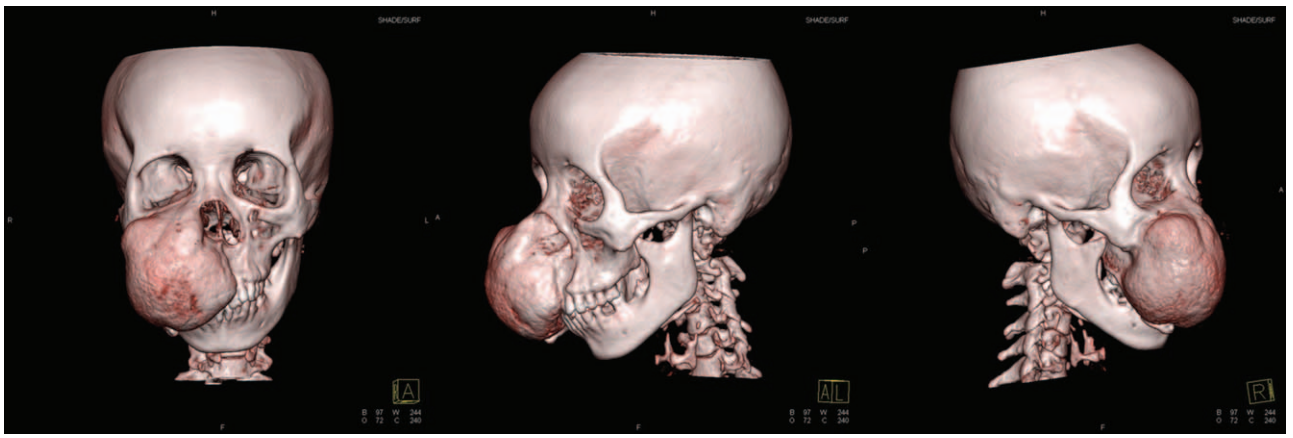

Figure 4. Three-dimensional reconstruction in different directions.

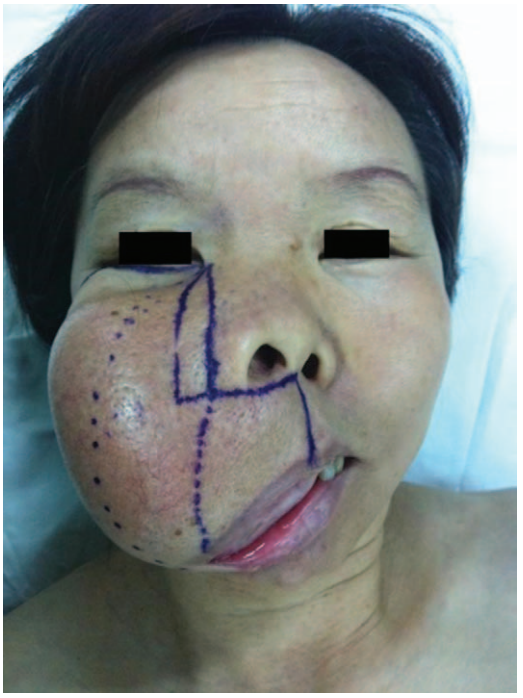

Figure 5. Preoperative labeling of primary excision.

OF be completely enucleated from the surrounding bone considering the high chance of recurrence.[1,18] When patients undergo extensive surgical resection, additional reconstruction using bone grafts or implants must be considered because of esthetic and functional problems.[19,20] Silveira et al[7] performed fixation using a reconstruction plate to completely remove the lesion in 1 patient. Tyagi et al[21] suggested that giant maxillary lesions should not be assumed to be aggressive, but should be carefully removed using thorough enucleation performed by curettage of the tumor cavity walls followed by prosthetic replacement for maxillary defects. Whether the tumor needs to be completely removed depends on the growth rate. Although OF usually grows slowly, individual juvenile variants can show

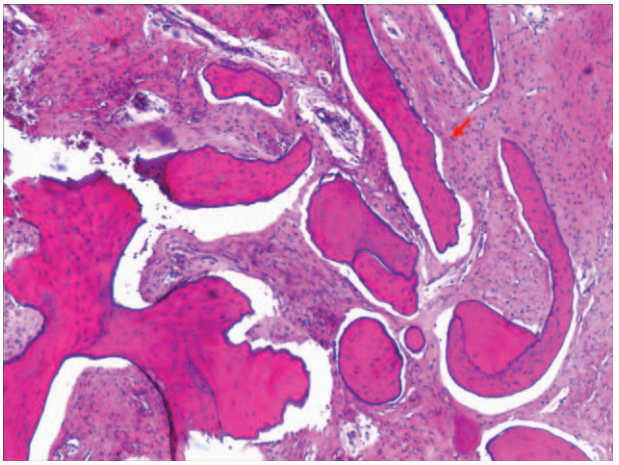

Figure 6. Microphotograph showed the trabecular bone around by osteoblast scattered within the proliferative fibrous tissue. Trabecular bone thickened and was arranged in parallel with hardening around which confirmed the diagnosis of ossifying fibroma (hematoxylin and eosin, ×40).

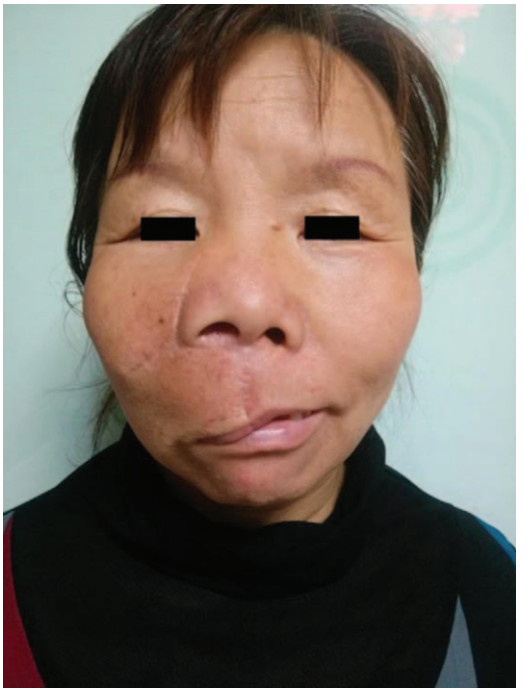

Figure 7. Two years after the primary excision, the patient complained of conspicuous scars and drooping of the upper lip on the right side with no visible increase in volume of the tumor.

aggressive growth ("juvenile aggressive OF").[15,22,23] Agarwal et al[24] reported a case of giant maxillary OF in a 15-year-old female patient with left eye protrusion that was treated by totally resecting the maxilla through the Weber–Ferguson approach. In our case, the giant lesion was extensive and involved the inferior

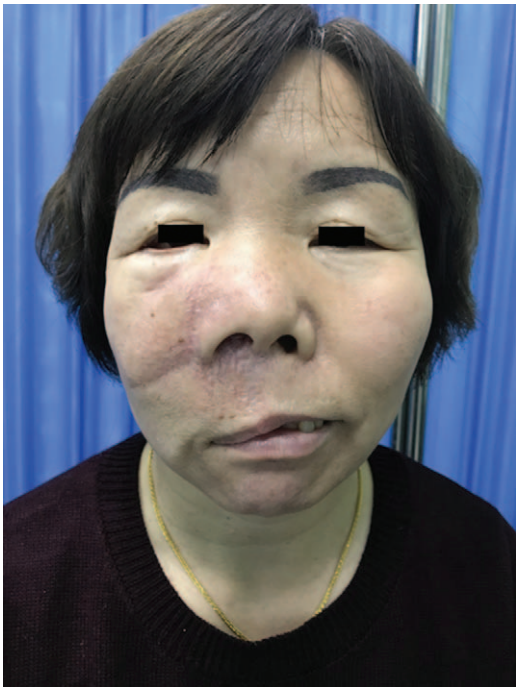

Figure 8. Fifteen months after the second surgery, local infection caused by bone wax rejection leads to subcutaneous adhesion of the right lower eyelid, which finally resulted in right lower eyelid ectropion through scar contracture.

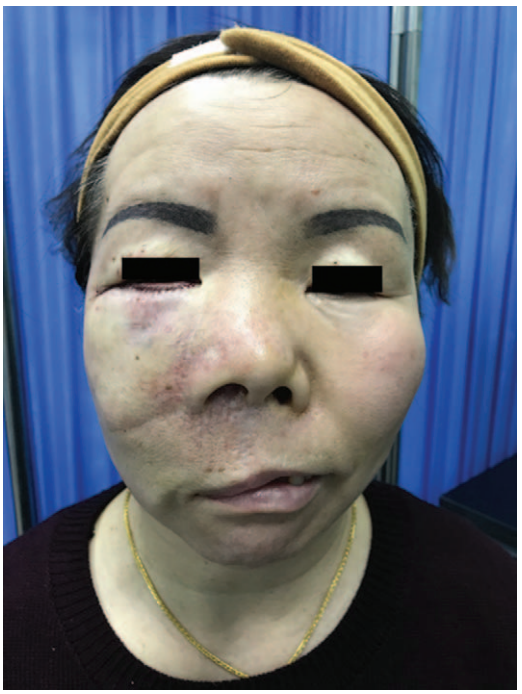

Figure 9. Postoperative photograph of the third surgery.

orbital wall, maxillary sinus, right os nasal, and right alveolar bone, and therefore required reconstruction to maintain the function of adjacent organs after complete resection. Since our patient refused bone or metal grafting for financial reasons with 30-year slow-growing mass, we performed conservative excision using the Weber–Fergusson approach, considering the esthetic and functional problems. We also followed the patient regularly postoperatively. Within the 4 years after the first operation, some complications occurred such as scar formation, scar contracture, and valgus deformity in the right lower eyelid. In order to improve the appearance of the face, we performed facial deformity correction in November 2015 to correct significant scar on the face and overhanging upper lip on the right side (Fig. 7). In February 2017, we corrected the right lower eyelid valgus caused by scar contracture through scar releasing and prosthesis implantation (Fig. 9). Following the third surgery in January 2018, the patient expressed satisfaction with a better facial appearance.

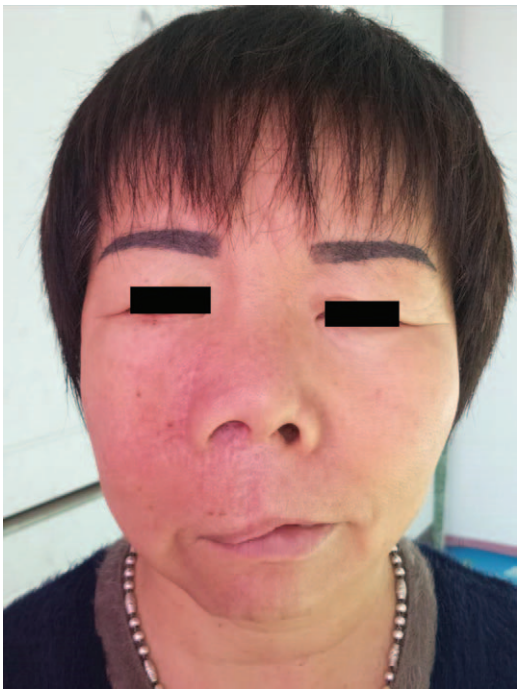

Figure 10. Eleven months after the third surgery.

Since we chose conservative partial excision, the follow-up of tumor growth is critical. Mintz and Velez[9] and Chang et al[4] described OF recurrence rates ranging from 30% to 58% and 0% to 28%, respectively, showing that recurrence rates vary and indicating that patients should be followed-up regularly. Previous studies stress the tumor recurrence rate and ignore follow-ups to assess facial appearance and determine corrective treatments. Because of the postoperative deformities that developed in our patient, including obvious depression at the resection site, facial asymmetry, and eventual scar contracture, we paid more attention to improving facial appearance. In our case, no visible increase in volume of the tumor was seen within 4 years after resecting the tumor, and the patient did not complain about the enlargement of the tumor again. With 2 surgical procedures to improve her facial appearance, the patient got a more confident psychological state and better quality of life. However, follow-up is still necessary to monitor for complications and tumor recurrence.

4. Conclusion

Giant maxillary OF is rare, and there is no reported uniform standardized treatment. When OF lesions are giant and invasion extensive, the growth rate of the tumor and whether facial organ function is affected should be considered to develop treatment programs individualized to the patient. Since lesions grow slowly, we suppose that it is feasible to excise conservatively when reconstruction cannot be operated considering the esthetic and functional problems. Also regular postoperative follow-up is necessary to detect recurrence, and to improve facial appearance as required.

Author contributions

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Validation: Lianbo Zhang.
Writing – original draft: Zhuoxia Wu.
Writing – review & editing: Zhuoxia Wu.

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