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Assessment of preoperative and postoperative quality of life of 24 patients with fibrous dysplasia of the mandible

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Abstract

The aims of this study were to compare preoperative and postoperative quality of life (QoL) in 24 patients with fibrous dysplasia of the mandible, and evaluate the effects of two surgical techniques on their postoperative QoL. Their QoL was assessed using the University of Washington Quality of Life (UW-QoL) questionnaire. The patients was divided into two groups according to the two different surgical techniques used. The first group (n = 11) were managed with focal bone modification, and their results compared with those of the other group (n = 13) who were managed with total resection of the focal bone. Their total postoperative QoL score of patients was significantly higher than that of the preoperative period ($p = 0.035$). The postoperative scores for activity ($p = 0.004$), recreation ($p < 0.001$), chewing ($p = 0.03$), and speech ($p = 0.001$) were significantly lower than those before operation, and those for pain ($p < 0.001$), appearance ($p < 0.001$), mood ($p < 0.001$), and anxiety ($p = 0.001$) were significantly higher. The change in scores for each patient (between before and after the operation) also differed. The UW-QOL can be used to evaluate the QoL of patients with fibrous dysplasia of the mandible, and operation can improve it. Different surgical techniques have a significant influence on patients' QoL.

Keywords: fibrous dysplasia of mandible; quality of life; University of Washington head and neck Quality-of-life

Introduction

Fibrous dysplasia is a rare, benign disease of bone, the main feature of which is that normal bone is replaced by fibrous tissue. In 1937, Albright et al[1] reported a disease that occurred in women with precocious puberty, the pathological features of which were similar to those of fibrous dysplasia, with accompanying skin pigmentation. They described it as disseminated fibrous osteitis and named it McCune-Albright syndrome. In 1938, Lichtenstein[2] described the disease for the first time and named it fibrous dysplasia. The disease can occur in any bony tissue, and half was in craniofacial bone.[3] It can be classified into "multiple bone" type, "single bone" type, and "McCune-Albright" type. In the "multiple bone" type, the craniofacial bone is affected in 50%–100% of patients. The incidence in the mandible is about half that in the maxilla, and the disease is usually found unilaterally in the mandible.

Most fibrous dysplasias develop slowly, the early oral and maxillofacial symptoms are not obvious, and they can easily be ignored by patients. As the disease progresses slowly, the symptoms such as occlusion disorder and loosening of teeth can develop. Severe dysplasia can result in maxillofacial deformity, it has a severe aesthetic impact, and it brings a certain psychological burden to patients. When the lesion is associated with infection, there is obvious, radiating pain. If it invades the nerve it can affect the vision and hearing, and cause a degree of facial paralysis. It may also result in nasal

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congestion if it invades the sinus cavity. When the lesion is large, it can cause pathological fractures of the maxillofacial skeleton, or the development of craniocerebral symptoms.

With the concept of "health-related survival", the meaning of quality of life (QoL) becomes more precise and perfect. The QoL of patients with fibrous dysplasia should also be emphasised, which is of great importance when it comes to the selection of treatment options. In this research, therefore, we have used the University of Washington QoL questionnaire to assess the patients' preoperative and postoperative QoL, and to assess the effects of different treatments on it.

Patients and methods

Patients

We collected 28 patients who had been diagnosed with fibrous dysplasia and who were operated on at the School of Stomatology, China Medical University, from December 2014 to August 2016. We excluded four cases (recurrence, n = 2, and kidney failure and lost to follow-up, n = 1 each). A total of 24 patients were therefore included in the study, all of whom had complete clinical data and follow-up.

Surgical techniques

We selected the surgical technique according to the patient's aesthetic requirements, degree of recurrence, and tumour boundaries. Eleven cases were treated by focal bone modification and 13 by total resection, the focal bone modification being a more conservative surgical treatment. In view of the appearance, we resected the focal bone and tried to protect normal bone and facial shape as much as possible. The total resection of the focal bone is a total or extended resection that aims to reduce recurrence as far as possible. When the boundary of the tumour was relatively clear, we selected total resection.

The general demographic information of patients is given in Table 1, and there were no significant differences in age, weight, size of tumour, or operating time. All the symptoms of fibrous dysplasia were limited to the oral and maxillofacial region, and the two groups were similarly affected, which made them comparable.

Informed consent was obtained from each participant, and ethics approval was granted by the China Medical University Ethics Review Board.

University of Washington Quality of Life scores (UW-QoL)

The UW-QoL head and neck questionnaire, fourth edition of the Chinese version, was used. The content includes physical and non-physical aspects of QoL, including 12 items: pain, appearance, activity, recreation, swallowing, chewing, speech, and the shoulder function, taste, saliva, mood, and anxiety. Each of the 12 questions has between three and five possible options that allow patients to describe their current functional state. The highest score, or "normal" function, is assigned 100 points, with 0 representing the lowest. The higher the score, the better the QoL. The results of the scale were calculated according to the total scores of 12 items, with a full score of 1200.

Methods

We handed out questionnaires to patients preoperatively and they completed them in time. We reviewed the patients by telephone six months postoperatively, and to ensure the consistency of the evaluation, we gave no explanations and hints.

Statistical analysis

We used IBM SPSS Statistics for Windows (version 19.0, IBM Corp) for the statistical analysis, and the independent samples t test and paired samples t test were used to assess the significance of differences. Probabilities of less than 0.05 were accepted as significant.

Results

Preoperative and postoperative QoL

Twenty-four patients filled in the questionnaires as required. When we compared UW-QoL scores of patients before and six months after operation, the postoperative scores for activities, recreation, chewing, speech were lower than before, and those for pain, appearance, mood, and anxiety were higher than before (Table 2). There were no significant differences in the scores for saliva, swallowing, shoulder function, and taste before and after the operation. The total QoL of the postoperative patients was higher than that in the preoperative patients (Table 2).

The changes in scores as a result of the operation

In the bony modification group the changes in total scores for pain, appearance, activities, recreation, speech, mood, and anxiety, differed significantly. The changes in the scores for pain, appearance, mood, anxiety, were better and those in scores for activities, recreation, and speech were worse. In the resection group, the changes in scores for pain, appearance, activities, recreation, chewing, speech, and mood differed significantly (Table 3). The changes in scores for pain, appearance, and mood were for the better, and those for recreation, chewing, and speech were worse.

Table 1
Characteristics of patients. Data are mean (SD) unless otherwise stated.

Variable	Focal modification (n = 11)	Focal resection (n = 13)	t	df	p value*
Male/female	2/9	4/9	–	–	–
Age (years)	41 (13)	40 (17)	−0.215	21.775	0.832
Weight (kg)	63 (11)	61 (17)	−0.312	20.88	0.759
Size of tumour (cm^2)	9.2 (3.5)	6.4 (6.2)	−1.343	22	0.193
Duration of operation (min)	94 (44)	82 (64)	−0.548	21.18	0589

* Independent samples t test.

Table 2
University of Washington Quality of Life scores before and after operation in 24 patients. Data are mean (SD).

University of Washington QoL item	Before operation	After operation	t	df	p value*
Pain	57.3 (11.6)	87.5 (12.8)	−8.576	46	<0.001
Appearance	49.0 (11.6)	74.0 (21.5)	−5.019	46	<0.001
Activities	80.2 (12.7)	66.7 (17.5)	3.060	46	0.004
Recreation	80.2 (12.7)	61.5 (16.5)	4.417	46	<0.001
Swallowing	90.4 (15.3)	82.1 (19.5)	1.637	46	0.108
Chewing	87.5 (22.1)	70.8 (29.2)	2.230	46	0.031
Speech	100 (0)	87.6 (16.3)	3.715	46	0.001
Shoulder	100 (0)	100 (0)	–	–	1.0
Taste	100 (0)	95.9 (11.1)	1.813	46	0.076
Saliva	82.1 (16.8)	83.5 (16.9)	−0.283	45.99	0.778
Mood	49.0 (21.5)	72.9 (12.6)	−4.716	46	<0.001
Anxiety	41.6 (20.5)	59.9 (14.1)	−3.610	46	0.001
Total	917.2 (51.0)	942.3 (24.2)	−2.178	46	0.035

* Independent samples t test.

Table 3
Effect of operation on patients' University of Washington Quality of Life scores for each variable (score after operation minus that before operation). Data are mean (SD).

University of Washington QoL item	Focal modification (n = 11)	t (df = 10)	p value*	Focal resection (n = 13)	t (df = 12)	p value*
Pain	27.3 (17.5)	−5.164	<0.001	32.7 (21.4)	−5.516	<0.001
Appearance	25.0 (19.4)	−4.282	0.002	25.0 (25.0)	−3.606	0.004
Activities	−13.6 (13.1)	3.464	0.006	−13.5 (19.4)	2.501	0.028
Recreation	−22.7 (20.8)	3.627	0.005	−15.4 (19.2)	2.889	0.014
Swallowing	−6 (19.9)	1.0	0.341	−10.2 (28.4)	1.3	0.218
Chewing	−13.6 (32.3)	1.399	0.192	−19.2 (25.3)	2.739	0.018
Speech	−15 (17.2)	2.887	0.016	−10.2 (15.9)	2.309	0.040
Shoulder function	0 (0)	0	1.0	0 (0)	0	1.0
Taste	−3 (9.9)	1.0	0.341	−5.1 (12.4)	1.477	0.165
Saliva	6 (19.9)	−1.0	0.341	−2.5 (25.1)	0.365	0.721
Mood	29.5 (18.8)	−5.221	<0.001	19.2 (29.1)	−2.379	0.035
Anxiety	18.5 (17.8)	−3.464	0.006	18.2 (35.5)	−1.845	0.090
Total	32.4 (45.8)	−2.342	0.041	19.0 (49.5)	−1.384	0.192

* Paired samples t test.

Discussion

The treatment of fibrous dysplasia includes drugs and operation, and operation is used in most cases. The choices are still controversial.[3] The reasons for the choice of operation are first that drugs can relieve the disease only temporarily, and cannot eliminate the lesion completely; secondly, conservative surgical treatment improves the patient's appearance, but the resection is not complete and the later effect is not clear; and, finally, that complete resection of the lesions may cause a degree of bony deficit and facial deformity. It can seriously affect patients' oral function and has a serious impact on the life of patients as well as causing a heavy psychological burden.

It is therefore important clinically to evaluate the changes in postoperative QoL in patients with maxillofacial fibrous dysplasia. Table 1 shows that operation can improve the QoL of these patients and improve the pain, appearance, mood, and anxiety scores. However, it can also reduce the scores for activities, recreation, chewing, and speech. Some authors[4,5] have reported that maxillofacial fibrous dysplasia may result in osteomyelitis and pain. However, the improvement that we found in symptoms of pain is likely to be because some or all of the infected bony lesions were resected, and the flushing and other measures help to alleviate the inflammatory pain at the same time as the lesion is removed. In addition the appearance scores were improved.

Most of the current research recommends conservative surgical treatment to improve appearance. The patients in

the bony modification group accepted conservative surgery. Some patients, who had agreed to total resection, did not have large bony or interosseous defects that could have caused facial deformity, so their postoperative appearance improved greatly. The postoperative activity and recreation scores decreased. This may have been because the patients were still in the recovery period, or because the size of the bony defect had a great psychological impact on the patients, making them unwilling to go out. Postoperative chewing and speech function also declined. Many studies have shown that the integrity and continuity of the mandible is indispensable to maintain oral function such as chewing and pronunciation. The jaw defect may therefore be the main reason that the chewing and speech functions declined. Mood and anxiety were relieved postoperatively, which showed that the operation had eased the patient's psychological pressure to a certain extent, because it can resolve pain and restore facial appearance.

In recent years rates of survival and cure have improved constantly as we pay more attention to the QoL of postoperative patients, and this research can guide clinical rehabilitation. However, the patients' assessment of their condition is still the main way of evaluating QoL. At present, our interest in the patients' QoL is mainly concentrated on patients with head and neck cancer,[6,7] and its investigation in patients with fibrous dysplasia of the mandible is negligible. Facial deformity and postoperative bony defects cause various disorders and have psychological impacts that affect the life of patients with fibrous dysplasia, so we applied the UW-QoL questionnaire to this group of patients. From our results we have learned that the operation is necessary and helpful for them.

In addition, we should increase postoperative health guidance and social support for these patients. We can also use new technology such as computer navigation to assist more accurate modification of the bone, and also insert prostheses to improve the shape of the face, as well as transplanting a fibular free flap to make chewing, swallowing, speech, breathing, movement, and feeling easier. These will significantly improve the physiological and psychological aspects of the QoL.

We also analysed the influence of two different surgical methods on the QoL of patients with fibrous dysplasia: bony modification and total or expanded excision. Patients can also have repair and reconstruction with a fibular free flap, iliac bone, or prosthesis if needed.[8–11] Some papers have reported that complications have arisen as the disease has developed, such as damage to the facial nerve leading to facial paralysis, and damage to the auditory nerve leading to nerve deafness. Fibrous dysplasia in the orbital area can also cause permanent impairment of vision, and severe cases can lead to blindness.

The expanded resection is the only way to ensure a radical cure.[12] The first method of surgical treatment is mainly to correct or prevent the development of functional deformity and improve the appearance of face, and the second is radical treatment, which minimises the possibility of recurrence. However, it can cause a defect or discontinuity of the jaw. The surgical method chosen is based on the patients' age, lesion site, size, and radiographic classification. Some authors think that asymptomatic mandibular fibrous dysplasia should be operated on after puberty,[13] but this needs further study. Numerous studies have shown that the area of the bony defect has an important role in postoperative QoL. Schliephake et al[14] reported that the bony defect left behind after mandibular excision, even if a free flap is used to repair it, would significantly affect the QoL. Sharma et al[12] considered that such a wide resection would not only greatly increase the difficulty of the operation, but also caused a lot of trauma to patients themselves, such as affecting chewing, swallowing, and other oral functions (as well as the beauty of the face) while reducing the recurrence rate at the same time. It could also affect the UW-QoL score.

In our study of 24 patients, 11 patients accepted focal bone modification or conservative surgery and 13 cases total focal osteotomy. Table 3 shows that the different operating techniques had different influences on QoL. In the scores for pain, appearance, and mood, the changes in the two groups were for the better. The two methods could both improve pain, appearance, and mood. In the scores for activities, recreation and speech, however, the changes were not good. The two methods both reduced activities, recreation, and speech. However, in the focal modification group there was no significant difference in chewing (suggesting that focal bone modification had less influence on chewing), so it may be related to the conservative nature of the resection. In the bony resection group there were no significant differences in anxiety or total score, which suggests that total resection of the focal bone does not improve anxiety and the total score like the bony modification did. It may be that the total or extended resection leaves more bony defects and therefore causes postoperative anxiety.

The shortcomings of our study are first that the follow-up period is too short, and secondly it is not a prospective, randomised, controlled trial. The short follow-up limits our observations both about recurrence and about the long-term implications of the different surgical techniques. The sample size of 24 may also limit the reliability of our results. We should increase the sample to achieve a more reliable conclusion in any further study.

We will review the patients in the focal bone modification group for a long time, and observe the changes in the disease. We will totally excise the focal bone when necessary. The indications for operation in these patients are controversial. According to our present result, the treatment of maxillofacial fibrous dysplasia should be more conservative, and we should attach more emphasis on improving patients' QoL.

We hope that the assessment of preoperative and postoperative QoL of patients with fibrous dysplasia and the assessment of different surgical techniques on their QoL

can contribute some suggestions for future improvements in treatment to both patients and surgeons.

We conclude that the operation can improve the quality of life of patients with fibrous dysplasia. Focal bone modification and total excision of focal bone have different influences on their QoL.

Conflict of interest

We have no conflicts of interest.

Ethics approval and consent to participate

Ethics approval was granted from the China Medical University Ethics Review Board. Informed consent was obtained from each participant.

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