

Surgical Management of Fibrous Dysplasia-Related Lip Paresthesia: a Case Report

SUMMARY

Background/Aim: This study aims to describe a case of fibrous dysplasia causing lip paresthesia and the surgical treatment applied, highlighting the roles of dentists and oral and maxillofacial surgeons in managing such cases. **Case Report:** A 37-year-old female presented with right-sided lip paresthesia. Diagnostic imaging, including panoramic radiography and Cone Beam Computed Tomography (CBCT), was used to assess the lesion. The patient underwent surgery under general anesthesia. After local anesthesia, an incision was made, the periosteum was elevated, and the expanded bone was exposed. The buccal cortical bone was removed, and the radiopaque lesion was enucleated, exposing the mandibular nerve. The excised tissue was sent for pathological examination. Postoperative care included antibiotics, painkillers, and mouthwash, with detailed wound care instructions. A follow-up visit was conducted one year post-surgery. Panoramic radiograph and CBCT revealed a radiopaque lesion above the mandibular canal with a radiolucent halo. The lesion was successfully enucleated, exposing the mandibular nerve without further damage. Histopathology confirmed fibrous dysplasia. One year post-surgery, the patient had no recurrence of paresthesia or lesion regrowth, although a slight radiolucent area remained visible on the panoramic x-ray. **Conclusions:** Fibrous dysplasia is a rare, benign bone disorder requiring comprehensive diagnostic evaluation and careful management. In this case, surgical excision was necessary due to the lesion's impact on the mandibular canal and resulting lip paresthesia. The successful outcome highlights the importance of early diagnosis, interdisciplinary collaboration, and long-term follow-up.

Key-words: Enucleation, Fibrous Dysplasia, Fibro-osseous Lesions, Lip Paresthesia, Maxillofacial Surgery

Atalay Elver, Bedriye Gizem Çelebioğlu Genç

Department of Oral and Maxillofacial Surgery,
Cyprus University of Health and Social Sciences,
Turkey

CASE REPORT (CR)

Balk J Dent Med, 2024;212-215

Introduction

Fibrous dysplasia represents a rare skeletal disorder characterized by pathological proliferation of fibrous tissue in place of normal bone, presenting as a benign fibroosseous lesion. It can manifest in two distinct forms: monostotic fibrous dysplasia, which affects a localized region and polyostotic fibrous dysplasia, characterized by a systemic involvement that may be accompanied by café-au-lait spots of skin pigmentation and hormonal

imbalances, constituting what is referred to as McCune-Albright syndrome¹⁻⁴.

The precise incidence of fibrous dysplasia remains somewhat uncertain, comprising approximately 2.5% of all bone tumors and nearly 7% of benign bone pathology². Treatment strategies for fibrous dysplasia of the jaw span a spectrum, ranging from vigilant observation to surgical intervention, contingent upon the progression of the condition. Early studies advocate for extensive surgical resection and immediate reconstruction of the affected

regions in the maxilla and mandible, considering the lack of anticipated recurrence⁵.

The condition is defined as of genetic origin, arising from activating mutations in the GNAS complex locus gene located on chromosome 20, culminating in somatic activating mutations of the G α subunit of a G protein-coupled receptor and subsequent upregulation of cyclic adenosine monophosphate⁶.

As fibrous dysplasia matures, it assumes a mixed radiolucent/radiopaque appearance, with established lesions displaying mottled radiopaque patterns reminiscent of “ground glass” characterized by small, diffusely dispersed opacities^{7,8}. Ongoing research endeavors continue to investigate the potential role of medical therapies in managing fibrous dysplasia. The utilization of bisphosphonates as a treatment strategy is founded on the rationale that fibrous dysplasia, akin to other osteoclast-driven disorders such as Paget’s disease of bone, exhibits increased osteoclastic bone resorption induced by interleukin-6. Various reports have demonstrated the significant reduction of bone pain and bone turnover with the use of pamidronic and alendronic acid⁹.

The main purpose of this study is to describe a case of fibrous dysplasia that causes lip paresthesia and the surgical method applied for its treatment. With this presentation, we emphasize the critical role of both dentists and oral and maxillofacial surgeons in managing complex cases such as this; it also gives information about the path to be followed. It also highlights the importance of long-term follow-up and keeping up with the latest researches in this field to ensure the best possible outcomes for patients.

Case Report

A 37-year-old female patient without any systemic disease applied to us with the complaint of right-sided lip paresthesia. In the panoramic radiograph taken from the patient, it was seen that there were no teeth in the right mandibular region, but there was a radiopaque lesion just above the mandibular canal with a radiolucent halo around it (Figure 1).



Figure 1. The first panoramic radiograph taken from the patient

A detailed history was taken from the patient, then an intraoral clinical examination was performed (Figure 2) and the patient was referred for CBCT for detailed examination (Figure 3). The patient was informed about the treatment plan and signed an informed consent.

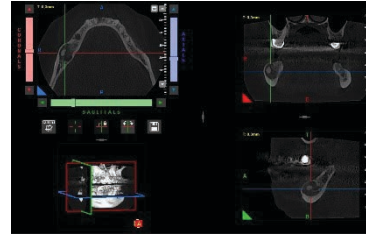


Figure 2. CBCT was used to determine the radiographic extent of the lesion and to reveal its relationship with anatomical structures

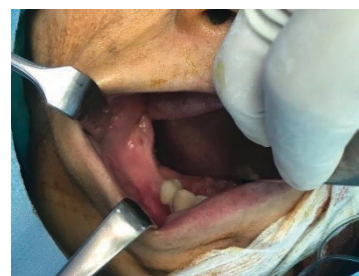


Figure 3. Intraoral view of the patient

The patient was operated under general anesthesia. After local anesthesia, an incision was made with a scalpel no. 15. After the periosteum was elevated, the expansion of the bone was seen more clearly (Figure 4). After removing the buccal cortical bone with the help of a bur, a lesion that appeared radiopaque on radiography was obtained (Figure 5). After the lesion was completely enucleated, it was observed that the mandibular nerve was exposed (Figure 6). The removed piece was sent to pathology. Surgical wound closure was performed with 3.0 vicryl sutures (Figure 7), promoting optimal wound healing and lowering the risk of infection. Removed piece was sent to pathology for examination and definitive diagnosis (Figure 8). The patient was given prophylactic antibiotics, painkillers and mouthwash and was informed about postoperative wound care.

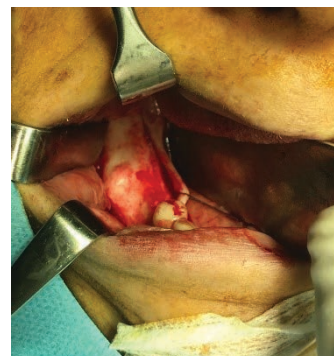


Figure 4. Expansion seen on the bone surface after elevating the periosteum

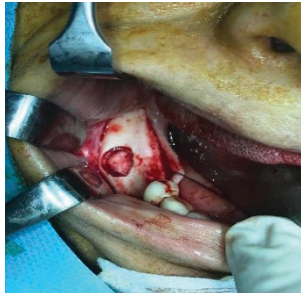


Figure 5. Fibrous-osseous tissue that appearing after removal the bone barrier



Figure 6. After completely enucleation was performed, we see that the mandibular nerve could be seen



Figure 7. Surgical wound closure was performed with 3.0 vicryl sutures



Figure 8. Appearance of the lesion excised and sent to pathology

The patient was called for a follow-up visit one year later, and the panoramic x-ray showed that the radiolucent area was still slightly visible. No paresthesia or recurrence was observed in the patient (Figure 9).



Figure 9. Postoperative radiograph a year after surgery

Discussion

Given the rarity of fibrous dysplasia cases, diagnosis requires a thorough radiological examination as well as a meticulous evaluation of the patient's medical history^{1,8}. With optimum preoperative preparation, precise surgical excision and careful postoperative monitoring, symptoms can be relieved and overall results can be improved in cases of maxillofacial fibrous dysplasia.

It is crucial to acknowledge that fibrous dysplasia, which frequently manifests in young individuals, can also develop in older age. The monostotic form is more prevalent and predominantly affects the 20 to 30 years age group, while polyostotic fibrous dysplasia primarily presents in children younger than 10 years of age^{3,4,7}. Our patient's case aligns with the literature data in this regard.

There is no definitive cure for fibrous dysplasia and the existing treatment guidelines lack universal consensus. Spontaneous resolution of fibrous dysplasia is exceedingly rare. As such, fibrous dysplastic lesions that remain asymptomatic, exhibit no progression and do not cause deformities or functional impairments should only be subject to regular monitoring. Surgical intervention becomes imperative when significant structures are at risk of compression^{2,7,8}. In our case, surgical operation was preferred because the lesion caused lower lip paresthesia on the patient's lip.

Patients should be reassured that fibrous dysplasia lesions are not malignant. Despite the low risk of recurrence and malignant transformation, long-term follow-up should not be neglected⁸. In the case of polyostotic fibrous dysplasia and adult patients, alendronate therapy is recommended to alleviate pain, improve bone turnover markers, and enhance bone scan findings⁹.

In particular, fibrous dysplasia lesions in the mandible can cause displacement of the mandibular canal, while those located in the maxilla can involve the maxillary sinus and potentially affect the base of the eye¹⁰. In our case, the lesion was compressing the mandibular canal, so surgical excision was preferred.

Conclusions

The first signs of fibrous dysplasia may be found incidentally by the dentist during a routine examination. For this reason, in addition to being able to recognize the characteristic findings of fibrous dysplasia, it is very important for dentists to guide the patient to determine whether there is systemic involvement and to work as a team with other colleagues.

References

1. Kruse A, Pieles U, Riener MO, Zunker Ch, Bredell MG, Grätz KW (2009). "Craniomaxillofacial fibrous dysplasia: a 10-year database 1996-2006". *Br J Oral Maxillofac Surg.* **47** (4): 302-305. doi: 10.1016/j.bjoms.2009.01.008. PMID: 19282072.
2. Obermeier KT, Hartung JT, Hildebrandt T, Dewenter I, Smolka W, Hesse E, et al. (2023). "Fibrous Dysplasia of the Jaw: Advances in Imaging and Treatment". *J Clin Med.* **12** (12): 4100. doi: 10.3390/jcm12124100. PMID: 37373793.
3. MacDonald DS (2015). "Maxillofacial fibro-osseous lesions". *Clin Radiol.* **70** (1): 25-36. doi: 10.1016/j.crad.2014.06.022. PMID: 25439185.
4. Pacino GA, Cocuzza S, Tonoli G, Boscolo Rizzo P, Tirelli G, Tofanelli M, et al. (2020). "Jawbone fibrous dysplasia: retrospective evaluation in a cases series surgically treated and short review of the literature". *Acta Biomed.* **92** (1): e2021018. doi: 10.23750/abm.v92i1.9904. PMID: 33682826.
5. Akashi M, Matsuo K, Shigeoka M, Kakei Y, Hasegawa T, Tachibana A, et al. (2017). "A Case Series of Fibro-Osseous Lesions of the Jaws". *Kobe J Med Sci.* 2017 **63** (3): E73-E79. PMID: 29434178.
6. Riminucci M, Robey PG, Saggio I, Bianco P (2010). "Skeletal progenitors and the GNAS gene: fibrous dysplasia of bone read through stem cells". *J Mol Endocrinol.* **45** (6): 355-364. doi: 10.1677/JME-10-0097. PMID: 20841428.
7. Feller L, Wood NH, Khammissa RA, Lemmer J, Raubenheimer EJ (2009). "The nature of fibrous dysplasia". *Head Face Med.* **5**: 22. doi: 10.1186/1746-160X-5-22. PMID: 19895712.
8. Burke AB, Collins MT, Boyce AM (2017). "Fibrous dysplasia of bone: craniofacial and dental implications". *Oral Dis.* **23** (6): 697-708. doi: 10.1111/odi.12563. PMID: 27493082.
9. Kitagawa Y, Tamai K, Ito H (2004). "Oral alendronate treatment for polyostotic fibrous dysplasia: a case report". *J Orthop Sci.* **9** (5): 521-525. doi: 10.1007/s00776-004-0809-0. PMID: 15449129.
10. Mortazavi H, Baharvand M, Safi Y, Behnaz M (2019). "Common conditions associated with displacement of the inferior alveolar nerve canal: A radiographic diagnostic aid". *Imaging Sci Dent.* **49** (2): 79-86. doi: 10.5624/isd.2019.49.2.79. PMID: 31281784.

Received on September 1, 2024.

Revised on September 10, 2024.

Accepted on September 17, 2024.

Conflict of Interests: Nothing to declare.

Financial Disclosure Statement: Nothing to declare.

Human Right Statement: All the procedures on humans were conducted in accordance with the Helsinki Declaration of 1975, as revised 2000. Consent was obtained from the patient/s and approved for the current study by national ethical committee.

Animal Rights Statement: None required.

Correspondence

Atalay Elver

Department of Oral and Maxillofacial Surgery

Cyprus University of Health and Social Sciences, Turkey

e-mail: atalayelver1@gmail.com