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CASE REPORT

Immediate allograft reconstruction of the infraorbital nerve following resection of polyostotic fibrous dysplasia lesion

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ABSTRACT

Processed nerve allografts (PNA) have increasingly been used as alternative to autogenous nerve grafts to repair nerve injuries in oral-maxillofacial surgeries. This case report describes an immediate PNA reconstruction of infraorbital nerve injury sustained during the ablation of a large expansile polyostotic fibrous dysplasia centered in the left maxilla.

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Polyostotic fibrous dysplasia; infraorbital nerve reconstruction; nerve allograft

Introduction

Fibrous dysplasia (FD) is an uncommon, benign skeletal disorder where normal bone is replaced with fibro-osseous tissue that can result in significant physical impairment [1]. FD arises due to postzygotic somatic activating mutations in the GNAS1 gene, which encodes the alpha subunit of the stimulatory G protein. Polyostotic fibrous dysplasia (PFD) is a form of FD affecting more than one skeletal site and comprises 15–30% of all FD cases [2,3]. PFD typically calls for conservative management such as bone contouring surgery due to the high rate of disease recurrence. However, in cases of PFD causing severe maxillofacial deformity, complete resection and reconstructive surgery may be considered [2,4].

Surgical management of pathology in the maxillofacial region often results in nerve injury. However, the decision to perform a nerve graft following an injury remains a controversial topic among head and neck surgeons [5]. For over five decades, the gold standard for reconstruction of nerve injury has remained the autogenous nerve graft [6–8]. However, the autogenous nerve technique can present with several clinical limitations including increased operating time for nerve graft harvest and postsurgical sequela at the donor site [9]. Recent studies have shown processed nerve allografts (PNAs) to be a safe and successful alternative to autogenous nerve grafts [10–16] with the additional benefit of avoiding donor site morbidity. Allogeneic nerve grafts are composed of decellularized and cleansed extracellular matrix from human cadaveric peripheral nerves. They are processed to be no immunogenic and inert, but able to retain the fascicular architecture and contain crucial molecules such as laminin for axonal guidance and outgrowth. These grafts are provided in different diameters and lengths to match reconstructive needs [5,10,13,15,16].

Most studies in the oral maxillofacial surgery literature involving PNAs have studied inferior alveolar and lingual nerve injuries. More recently, applications have emerged in the immediate inferior alveolar nerve reconstruction after ablative surgery of the mandible with 85–90% of patients achieving meaningful functional sensory recovery [13,15,16]. In addition to nerve sensory recovery, the functional motor recovery with PNAs for transected major peripheral nerves have also been reported with a 73% success rate and no related adverse events [12]. As a result of recent steps made in reducing the immunogenicity of nerve allografts, PNAs have become one of the most commonly used nerve allografts in the reconstruction of peripheral

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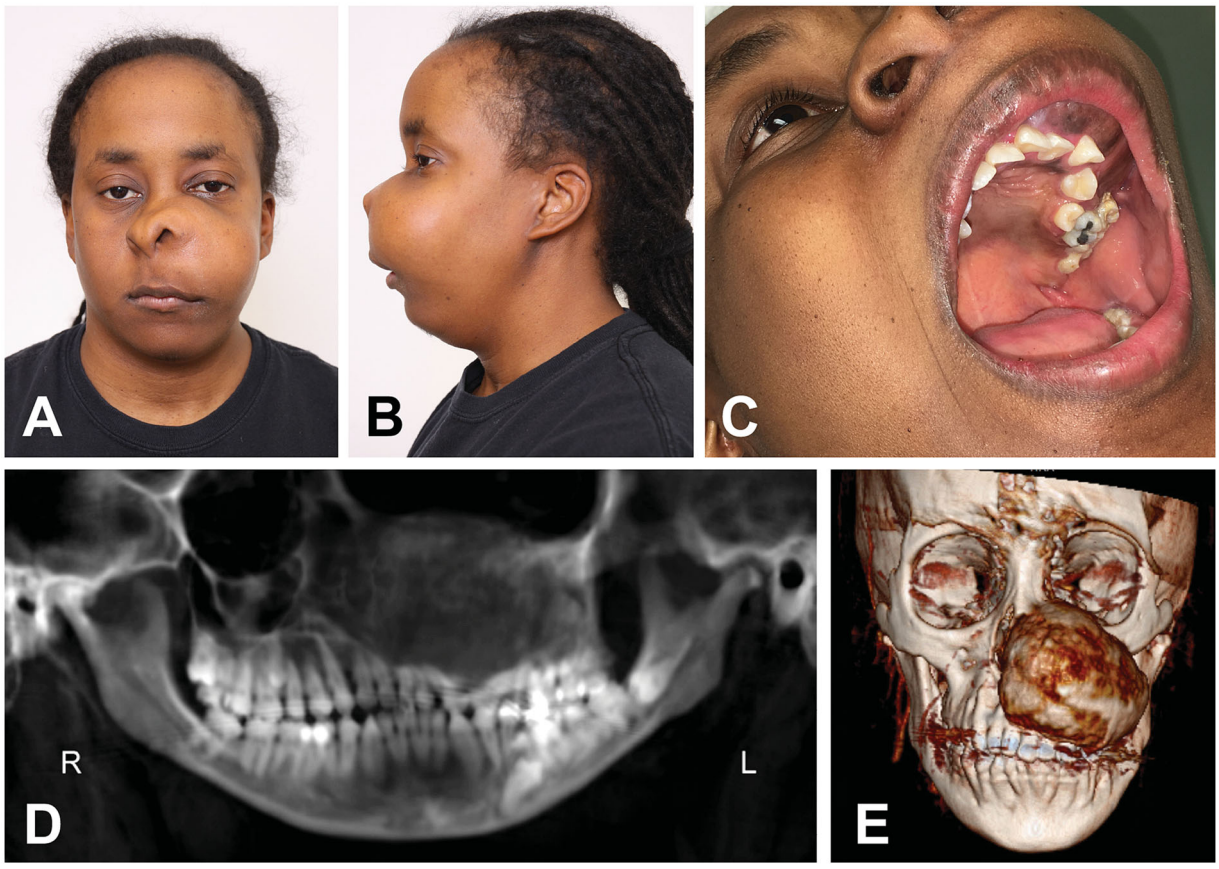

Figure 1. Preoperative images illustrating the severity and asymmetry of the maxillofacial deformity by the polyostotic fibrous dysplasia (PFD) lesion. (A-B) Frontal and lateral view of a large expansile mass of the left midface region causing ipsilateral nostril displacement. (C) Intraoral assessment demonstrates displacement of the left maxillary dentition. (D) Panoramic radiography shows the PFD lesion extending to the orbital floor, nasal region, maxillary and left maxillary dentition. (E) Cone beam computed tomography three-dimension (CBCT 3 D) reconstruction scan confirms the panoramic two-dimension findings.

nerve damage [10,12]. To the best of our knowledge, the technique and successful use of PNA in the immediate reconstruction of a resected infraorbital nerve have not yet been reported. This case report highlights the use of PNAs as a promising option for reconstructing the infraorbital nerve injury sustained during ablation of a large benign tumor. The principles of the Declaration of Helsinki and the Health Insurance Portability and Accountability were followed in this study. Informed consent was obtained from the patient.

Case report

A 27-year-old African American female that was referred to our institution for evaluation and management of a painless progressively enlarging mass in the left-sided midfacial region leading to severe facial deformity and asymmetry (Figure 1(A,B)). There was no history of trauma. Seven years earlier, this midfacial lesion was diagnosed as polyostotic fibrous dysplasia (PFD) requiring two previous facial bone contouring surgeries in 2012 and 2013 at an outside hospital. Her dental history included dental caries, xerostomia, stomatitis, and hyperdontia. She was taking trazodone 50 mg daily for underlying depression.

Intraoral examination showed the displacement of teeth due to extension of the PFD lesion (Figure 1(C)). Physical examination revealed fully preserved sensation in the left infraorbital nerve distribution with complete left and partial right nasal obstruction. Panoramic radiography and cone beam computed tomography three-dimension (CBCT 3 D) reconstruction scan demonstrated a lesion extending to the orbital floor, nasal region, maxillary and upper left dentition (Figure 1(D,E), respectively). Due to the severity and asymmetry of the maxillofacial deformity, a surgical approach was elected to complete removal of the PFD lesion including left hemi-maxillectomy and infraorbital nerve reconstruction with PNA (Avance®, AxoGen, Alachua, FL) followed by a complex 3 D reconstruction of the left hemi-maxilla and inferior orbital rim defect with left free fibula flap.

The midface degloving approach with subciliary and palatal sulcular incisions on the left maxilla was

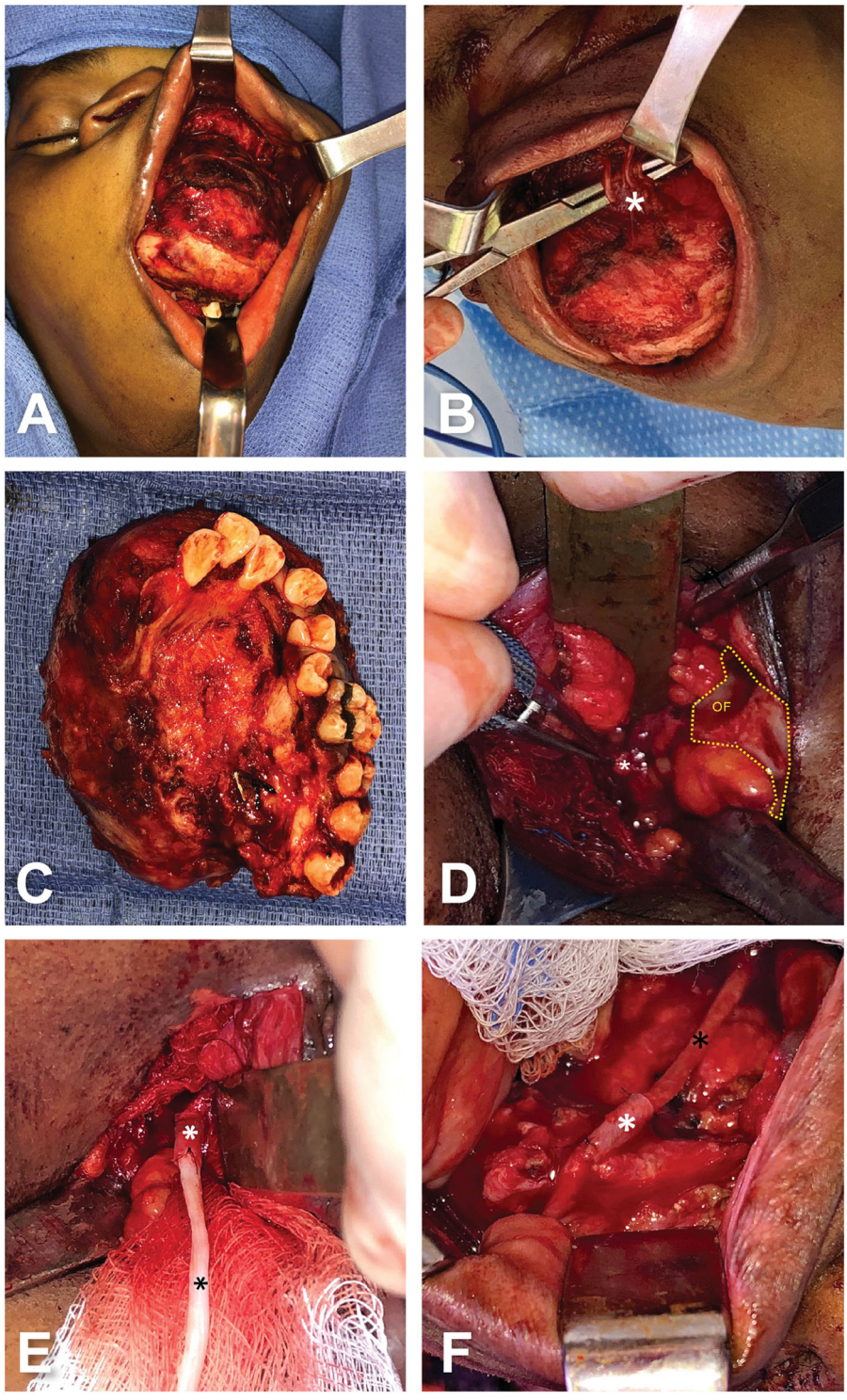

Figure 2. Intraoperative photographs illustrating the immediate reconstruction of resected infraorbital nerve using PNA. External photographs demonstrate the use of midface degloving approach. (A) Applying left subciliary and palatal sulcular incisions along the left maxillary region. (B) Infraorbital nerve (white asterisk) through the PFD lesion. (C) Specimen of left hemimaxillectomy. (D) Isolation of the infraorbital nerve stump (white asterisk) and orbital floor (yellow dotted area). (E) Coaptation of the PNA (black asterisk) to the proximal infraorbital nerve stump using 8-0 nylon suture followed by application of a nerve connector (white asterisk). (F) Similar procedure was performed at the distal infraorbital nerve stump.

used to gain access to the tumor (Figure 2(A)). Soft tissue exploration revealed a compressed infraorbital nerve situated within the PFD tumor (Figure 2(B)) resulting in the need for infraorbital nerve resection. The PFD lesion was completely removed after which was deemed to be a 90-mm tumor in the anterior-

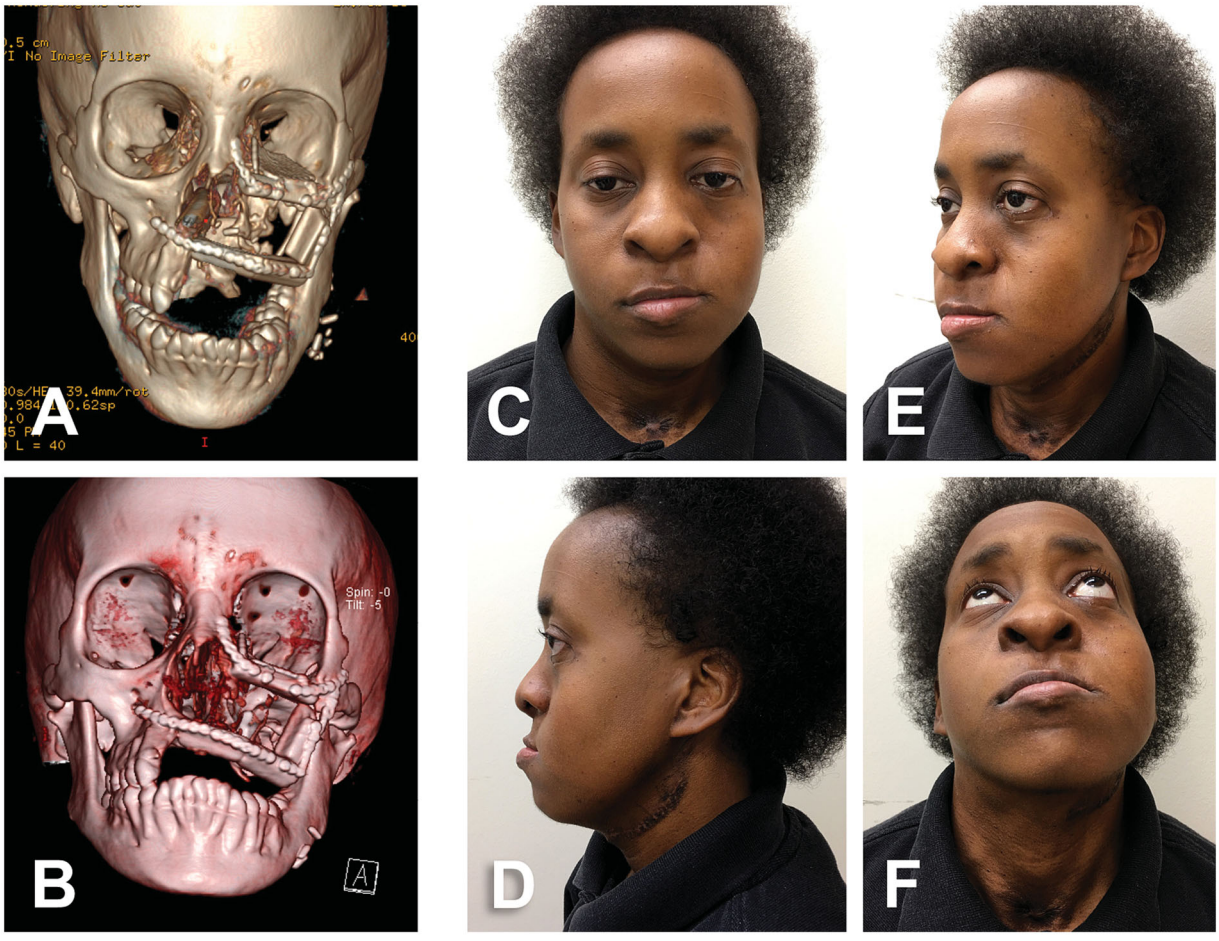

Figure 3. Images of postoperative assessment. (A) Immediate postoperative CBCT 3 D imaging scan of the left hemi-maxilla and inferior orbital rim reconstruction with free fibula flap following the removal of a benign lesion. (B) CBCT 3 D imaging scan at three-month follow up showing reconstruction plates and screws in place and without changes compared to that in the CBCT scan taken immediately after surgery. (C-F) Clinical photographs (six-month follow up) showing significant and stable facial symmetry.

posterior plane (Figure 2(C)). After the left hemi-maxillectomy was performed, as part of the PFD lesion removal, the infraorbital nerve was found to have a 70-mm defect in the superior-inferior plane. The proximal stump of the infraorbital nerve was isolated from surrounding tissues and marked with a 2-0 silk suture (Figure 2(D)), and subsequently its end was trimmed in preparation for the nerve repair. The same approach was done with the distal stump of the infraorbital nerve. Thereafter, the ends of a PNA (3-mm in diameter and 70-mm in length) were coapted to the proximal and distal infraorbital nerve ends with interrupted stitches of 8-0 nylon suture. Finally, nerve connectors were used around the coaptation sites to assist with the repair technique (Figure 2(E,F), respectively).

The patient immediately underwent to a complex 3 D reconstruction of the left hemi-maxilla and inferior orbital rim with 4-bony segments of left fibula free flap (Figure 3(A)).

Three months after surgery, the patient reached a significant facial symmetry supported by a stable underlying bony structure (Figure 3(A–F)). Five months after surgery, the patient recovered sensation in the left infraorbital nerve distribution. She exhibited static two-point discrimination with blunt-pointed tip at 15-mm, good directional discrimination over left nasal dorsum and left malar region. This assessment was graded as S3 on the Medical Research Council Scale [13]. This infraorbital nerve sensation has remained stable for the last few months and the patient is satisfied with the result.

Discussion

Polyostotic fibrous dysplasia (PFD) of the maxillofacial region can result in significant functional and aesthetics impairment. In this report, we described a patient with PFD, a benign bone disorder of unknown causes, that commonly affects the zygomatic, maxillary, and

mandibular region of the face [2]. Unlike the monostotic type, the polyostotic type is more likely to continue growing even after adolescence [2,17]. Thus, due to the severity of maxillofacial deformity and the relapse of previous facial bone contouring surgeries, our patient was planned to have a complete resection of the lesion followed by an immediate 3 D reconstruction surgery.

Neurosensory nerve injury following maxillofacial surgery can negatively impact both patients' functional and emotional quality of life. Even though neurosensory nerve damage is a common complication that occurs following ablative surgery of benign lesions, nerve repair is still not a routine procedure [5]. The low frequency of this surgical practice partly stems from beliefs that the risks outweigh the benefits of nerve repair using autogenous graft. Accordingly, a low number of surgeons routinely use nerve allografts or autografts for inferior alveolar nerve reconstruction following the resection of benign mandibular lesions [5].

Our case of infraorbital nerve reconstruction with PNA is consistent with other publications regarding PNA safety and successful outcome (85–90%) in nerve gaps of up to 70-mm in other locations [10,13–15]. Thus, our patient exhibited a clinically significant recovery of sensation at the 3-, 6-, and 12-month follow-ups. The reason why certain individuals are not able to regain some degree of sensory function after PNA reconstruction remains unclear. However, it seems likely that ongoing underlying inflammatory responses and pre-existing risk factors predispose some patients to an unfavorable outcome [10,12,13,16].

In conclusion, the real role of PNAs in nerve repair and their impact in the patients' quality of life remain to be further elucidated. Our case report demonstrates that PNA is a promising substitute for infraorbital nerve repair in the presence of large nerve gap following ablation of a benign tumor. Future cohort studies are needed to investigate whether PNAs are also an appropriate alternative for malignant lesions requiring postoperative chemotherapy and/or radiation therapy.

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Disclosure statement

Dr. Velasco Martinez is a consultant for Axogen Inc, Alachua, FL. Other authors have no financial disclosures.

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