GIANT PSAMMOMATOID OSSIFYING FIBROMA: MANAGEMENT CHALLENGES AND ROLE OF VIRTUAL SURGICAL PLANNING

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Abstract

Ossifying fibroma is a fibro-osseous tumor that tends to be well-defined, has a propensity for the mandible, and has a high potential for recurrence. Psammomatoid ossifying fibroma is an aggressive variant of juvenile ossifying fibroma and can destroy surrounding structures. This case describes the unusual presentation of psammomatoid ossifying fibroma of the mandible. A 30-year-old female patient presented with a history of progressive swelling on the right side of her face from the past 10 years, causing facial contour deformity. It details the diagnostic process, treatment challenges, and potential implications of a massive psammomatoid ossifying fibroma affecting the mandibular ramus. The clinical, radiological, and histological findings about management plans and outcomes are discussed and pertinent literature has been reviewed. The impact of the multidisciplinary approach on treatment outcomes and patient quality of life will also be taken into account. The worth of immediate reconstruction with free flaps and a 3D stereolithographic model is also discussed.

Keywords: Psammommatoid ossifying fibroma, Virtual surgical planning, Multidisciplinary, Reconstruction, Disarticulation resection

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Ossifying fibroma is a fibro-osseous lesion. Fibro osseous lesions are a set of diseases that are distinguished by the replacement of healthy bone with cellular fibrous tissue and a varying quantity of bone and cementum-like structures. The fibro-osseous lesions have three subtypes: ossifying fibroma, fibrous dysplasia, and cemento-ossifying fibroma. In 2005, the WHO designated all fibro-osseous lesions as bone diseases.[1]

Ossifying fibroma is a neoplastic, osteo pathology. There are two forms of it; Conventional ossifying fibroma and Juvenile ossifying fibroma. Conventional ossifying fibromas behave less aggressively. Whereas, Juvenile ossifying fibromas are more aggressive and more likely to recur. Compared to the conventional ossifying fibroma, which is more common in older people and the mandible, Juvenile ossifying fibroma tends to affect young adults more frequently and prefers the maxilla and paranasal sinuses.[2] Psammomatoid and trabecular ossifying fibromas are the two histologic variants of JOF.

Psammomatoid ossifying fibroma is more likely to develop in young adults, with the majority of cases occurring in the third to fourth decade. It has an equal predilection for both genders. Although it appears to be an innocuous swelling, depending on its size, location, and aggressiveness, it may damage nearby structures. There have been cases where psammomatoid ossifying fibromas have been associated with loss of vision, involvement of the base of the skull, and facial contour deformities.[3] Psomma-like bodies characterize this lesion, histopathologically. However, pathologists and clinicians have not come to a consensus on a clear criterion to distinguish between subtypes of ossifying fibroma due to complex histology and diverse clinical behavior. It often manifests radiologically as a radiolucent lesion with varying degrees of calcification. It has a characteristic ground glass appearance on plain

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radiography.[4] The correlation between clinicopathological and radiological findings is crucial for diagnosis. Treatment for POF is based on multiple factors: which include, location, duration, clinical symptoms, patient's general health condition, and lesion size. Treatment options are ranged from enucleation and curettage to radical excision. Many maxillofacial clinics worldwide, however, suggest radical excision with 0.5 cm safe margins as a treatment choice. POF has a recurrence rate that ranges from 30% to 50%. [1]

The use of minimally invasive surgery and multidisciplinary approaches in the management of POF is not sufficiently discussed in the literature. Furthermore, this case report emphasizes the importance of surgical planning using 3D stereolithographic models and immediate reconstruction, which has received less attention in earlier research.

Case Presentation

A 40-year-old female presented to us with a complaint of swelling on the right side of her face. The patient was otherwise healthy and had this swelling for the past 10 years. It was initially a small-sized swelling on the right lower jaw and increased in size progressively. There was no discomfort, fever, pus discharge, tooth mobility, missing tooth, or any relevant history. The patient denied having a history of such lesions in other parts of her body. Past medical, surgical, and family history was insignificant and had no association with this swelling. The patient had a below-average socioeconomic status, which was one of the prime reasons behind the treatment delay along with other contributing variables like anxiety over the diagnosis, emotionally distressing external factors, and lack of family support.

The patient had a characteristic anemic appearance. A thorough extra-oral examination revealed a deformed facial contour caused by a 15×10 cm swelling on the right side of the face, extending from the right infraorbital region to the right body region of the mandible in a superior-inferior dimension and transversely from the ramus region to the symphysis of the mandible. The swelling was obliterating the right nasolabial fold. It was also seen that the swelling had elevated the right ear lobe and had extended to the post-auricular region. Some of the portions of swelling particularly the inferior part had prominent vascular markings. On palpation, a firm, non-tender mass was noticed which was neither compressible nor reducible. The overlying skin was normal in color, temperature, and texture. Bilateral eye and neurosensory examination were normal. A thorough TMJ examination was not performed since the lesion had covered the right pre-auricular area. However, jaw movement and function were intact, suggesting a reasonably functional TMJ. Right-side facial nerve assessment (zygomatic, buccal, marginal mandibular branch) was similarly not possible.

The intraoral examination revealed cortical expansion of the right side of the mandible in the molar and premolar regions, extending to the ramus posteriorly and the symphysis anteriorly. Multiple carious teeth were present. However, none of the teeth was mobile. The overlying mucosa was intact. The mouth opening was satisfactory. Moreover, the aspiration of the lesion was negative.

An incisional biopsy was taken intraorally which revealed neoplastic tissue composed of numerous psammomatoid bodies embedded in the cellular fibrous stroma with occasional bony trabeculae, stroma was loose to densely cellular. The giant cell was also present within the stroma. A digital OPG was first performed, and it showed a sizable radiolucent-radiopaque osteolytic lesion extending from the right sub condylar region to the mandibular symphysis. A 3D reconstruction CT scan of the face was performed for surgical planning and diagnostic purposes. Which revealed an expansible lesion that was multilocular, mixed density, causing destruction of cortices and infiltration of the right side of the body and the ramus of the mandible, sparing the condylar processes. The body of the lesion showed a relatively homogeneous ground glass, but subtly variegated radio-dense appearance. The diagnosis of psammomatoid ossifying fibroma was made based on the correlation between the clinical, radiological, and histopathological data.

Resection of the tumor with 0.5 cm safe margins and disarticulation of the condyle was planned after extensive and critical discussion of the case with the

multidisciplinary team. This was followed by fixation with a prebent reconstruction plate and coverage of the intraoral defect with an anterior-lateral thigh flap. A preoperative tracheostomy was planned to prevent postoperative edema caused by venous congestion after free flap reconstruction. Post-operative ICU care and nutritional support were also recommended by the team. A replica with a stereolithographic model was created using CAD/CAM technology and a pre-operative CT scan. This replicated model was used for preoperative virtual surgical planning to determine resection margins and minimally invasive surgical approaches. The tumor resection was computer simulated to determine the precise placement of osteotomy cuts. The defect was reconstructed using the three-dimensional mirror image of the contralateral unaffected side. The reconstruction plate was bent using the reconstructed model as a template to rehabilitate the facial contour.

The patient received a transfusion of two pints of whole blood to treat her hemoglobin deficiency. The disease's aggressiveness, potential side effects, available treatment options, and complications were explained to the patient and her family. An informed consent form was filled out, along with a special consent form for the disarticulation of the TMJ and possible damage to the facial nerve.

Following deep general anesthesia and standard aseptic measures, a modified Blaire incision with its cervical extension continuing with the lower lip split incision was made. The lip split incision was included in the treatment plan considering the extent of the lesion and the need for adequate exposure. A flap was carefully reflected to approach the tumor and the remaining mandible. The overlying skin was extremely thin, so to overcome buttonhole formation blunt finger dissection was done. Osteotomy cuts were made in accordance with virtual surgical planning, and the condyle was disarticulated to completely remove the tumor. To ensure disease free tissue bed for free flap reconstruction, marrow frozen sections were sent, which came out to be negative for any pathological condition. There was significant intraoperative bleeding owing to the high vascularity of the tumor. The main branches of the facial nerve were preserved and the condyle was disarticulated. The pre-bended titanium reconstruction plate was fixed with three screws on the symphysial side of the defect and was stitched to remnants of TMJ on the proximal end. The surgical defect and reconstruction plate were covered by an anterolateral thigh flap. Maxillomandibular fixation was done with eyelets wiring, to aid osteointegration of the reconstruction plate. Post-operative ICU care was provided to the patient. The post-operative patient care plan included provisions for tracheostomy care, surgical wound management, routine flap monitoring, and nutritional support. Overall, The patient showed good recovery post-operatively. Marked post-operative edema of the lower face and neck was noticed, which was managed by postoperative antibiotics and steroids. The retromandibular part of the incision was dehisced, which was managed by regular normal saline dressing. The patient developed psychosis during her 1st postoperative week, after consultation with the psychiatry department, oral amitryptiline was prescribed (once daily at night for one month). Mild facial nerve weakness of the right temporal and zygomatic branches was observed. The patient was followed up for 2 years and showed no recurrence or any other late complication of treatment. Facial nerve functions were adequately preserved. The flap was fully taken by the recipient site. Moreover, the patient was satisfied with her treatment outcomes in terms of aesthetics and function. However, she had complained of the deviation of the mandible on the ipsilateral side, while opening the jaw. Other complaints include numbness of the lower lip and right half tongue. Both of the issues were inevitable disease related outcomes, and the patient was reassured and prior-counseled.

DISCUSSION

The fibro-osseous lesions are distinguished by a genetically controlled substitution of the normal bone architecture with fibro-osseous tissue.[5] The biological characteristics of fibro-osseous lesions are diverse, ranging from self-limiting hamartomas to aggressive

benign tumors. A definitive diagnosis is inevitable to devise an effective treatment plan. Ossifying fibroma is one of the rare, however clinically challenging fibro-osseous lesions affecting the jaws.

Ossifying fibroma is a benign tumor, attributed to bony expansion, local destruction, and recurrent behavior. It is broadly classified into two forms: conventional and juvenile ossifying fibroma. In juvenile ossifying fibroma, two histopathological variants have been identified: Trabecular type and psammomatoid type.[6] Osteonectin, a bone-specific protein, is detected positively by immunohistochemical (IHC) analysis, indicating that the tumor has an osseous origin.[7] JOF is believed to develop from the mesenchymal cells in periodontal ligaments, which serve as the precursor cells for cementum, fibrous tissue, and bone.[8] Additionally, these lesions have been shown to harbor a mutation in the tumor precursor gene HRPT2.[9] A rearrangement of the long arm of chromosome 12 that includes the genes MDM2 and RASAL1 may be responsible for juvenile ossifying fibroma and may serve as a diagnostic and predictive marker for aggressive behavior.[10]

JPOF is primarily a craniofacial pathology with only 10% of cases affecting the mandible, whereas the conventional; ossifying fibroma occurs frequently in the mandible. Juvenile ossifying fibroma may therefore be distinguished from conventional ossifying fibroma based on the patient's age, anatomical region of involvement, high propensity for recurrence, and aggressive behavior. Juvenile ossifying fibroma is reported to have a high incidence in children under the age of 15 years[11]. Owing to its rarity, No prevalence statistics are available for JPOF.[1] Compared to the psammomatoid variation (16–33 years)., the trabecular variety affects people who are younger (8–12 years).[12] The case we present demonstrates the presence of psammomatoid ossifying fibroma in the mandible at a later age, which is unusual in the literature. Based on a thorough study of the local, regional, and worldwide literature, this case can be included among the humongous JPOFs of the mandible that have been documented to date. The occurrence of JOF in older ages is an exception. Therefore, diagnostic criteria in these cases should be applied strictly, to avoid misdiagnosing the particular case with conventional ossifying fibroma.[4] JPOF has an equal predilection for both genders.[13]

Clinically, psammomatoid ossifying fibroma is asymptomatic unless the bone cortices expand. Typically, the patient's chief complaint is an expanding jaw swelling resulting in facial deformity.[14,12] Although PJOF is often painless, nerve compression can cause neuralgia-related facial pain disorder. Signs and symptoms of JPOFs are primarily governed by the location of the lesion and invasion of adjacent anatomical landmarks. Maxillary and paranasal sinus tumors, which constitute 85% of the PJOF, can cause blockage of the nose, and loss of vision, and result in proptosis of the eye due to optic nerve compression and displacement of orbital contents[1]. Whereas, skull base tumors constitute 3% of all the PJOF and usually present as intractable headaches and other neurological signs.[3] In our case, the patient reported swelling on the right preauricular area over the previous 10 years; it was three-dimensionally extending to the mid-cheek region, infra-orbital region, and first neck crease. The swelling was not associated with any functional concern or other significant findings.

Three phases of juvenile ossifying fibroma may be distinguished radiographically.[12] Stage I is characterized by a well-defined radiolucent lesion. Stage II radiolucent lesion with regions of interior calcification, the mixed stage. Stage III is the mature phase, which is attributed to fully radio-opaque bulk.[12,15] In CT, three patterns have been identified. First pattern: A thick outer mantle encircling a radiolucent inner core. The second pattern is the ground glass mural nodule. The homogeneous solid radiodense lesion is the third common pattern, in contrast.[16] Ground-glass opacity is not a JPOF-specific property. It is also associated with some other pathological conditions like fibrous dysplasia. Because the two conditions have different therapeutic implications, it is crucial to distinguish JPOF from fibrous dysplasia. As opposed to JPOF, which has a boundary that is clearly defined, fibrous dysplasia has a diffuse border that imperceptibly fades into the surrounding normal bone.[4] The ground glass

opacity is absent in JTOF. The interior structure is instead mostly radiolucent with sporadic and uneven calcifications.[4,17] Aneurysmal bone cyst formation is found in conjugation with psammomatoid juvenile ossifying fibroma, commonly in younger patients with an aggressive nature and high recurrence rate.[12] CT scan findings in this patient's case were consistent with stage II, mixed phase. And had a typical ground glass mural nodule with an outer mantle and sporadic calcifications.

The histopathological presentation of abundant basophilic calcifications in a fibrous stroma consisting of a central basophilic region encircled by peripheral eosinophilic boundary resembling psammomatoid bodies pathognomic of PJOF.[1] We made a conclusive diagnosis of PJOF based on histopathological evidence of psammamotoid bodies in the gross specimen and biopsied tissue. The differential diagnosis for JOF includes fibrous dysplasia, aneurysmal bone cyst, osteosarcoma, odontoma, ameloblastoma, osteoblastoma, and cement-osseous dysplasia.[1,14,18] Although the literature has not addressed the significance of the intraoperative marrow frozen section, our experiences have shown that it is a useful laboratory tool considering the high likelihood of disease recurrence.

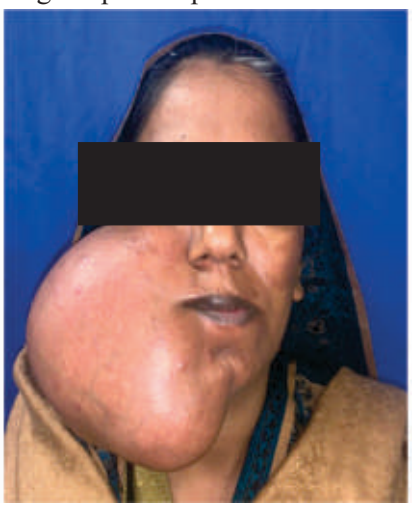
There is debate about whether aggressive treatment, such as local radical surgery, is better than conservative treatment, such as total excision, enucleation, or partial resection, which aims to keep the facial deformity to a minimum while preserving normal appearance and function.[1,19,14] Tanaka et al. and Leimola-Virtanen et al. both advocate for a more conservative approach to treating JOF, in the pediatric population. Contrarily, Toro et al., El-Mofty et al., Smith et al., Sun et al., and Partridges et al. preferred a more radical course of action, including en bloc resection with rigid internal fixation by reconstruction plates, bone grafts, integrated bone implants, and prosthetic rehabilitation.[20] Kaban et al. advocated curettage or enucleation in combination with adjuvant interferon-alpha therapy for 6 to 8 months as an alternate treatment for children with aggressive giant cell tumors.[20] In a pediatric population, cryotherapy, combined with enucleation, is considered a conservative treatment choice. However, chemotherapy and radiotherapy play no role in the treatment of PJOF. Conservative therapy is associated with a high rate of recurrence, even though it allows unrestricted growth of jaws.[1,15,20] Disarticulation resection of the mandible is an uncommon variant of segmental mandibular resection. A variety of pathologic diseases of the jaws and adjacent structures necessitate disarticulation resection. Many cases of PJOF have been reported in which disarticulation resection was performed.[1,21,22] The reconstruction of deformities associated with condylar disarticulation is complex and multistaged and may result in significant facial deformity and loss of mandibular function. Free fibula flap has become a standard treatment option for such defects.[23]

To treat this massive PJOF, an aggressive surgical technique was used. Three-dimensional virtual planning using a stereolithographic model and navigation system substantially benefited us in the decision-making process and the design of osteotomies. Because the modified Blaire incision and its cervical extension were insufficient to reach the tumor, a lower lip-split incision was used. This approach reduced operational complexity and improved access to the tumor and anatomical components. The postoperative aesthetic outcomes were likewise positive. When there is a possibility of tearing underlying skin flaps, delicate finger dissection is advised. As the remaining condylar stump in our case was less than 1 cm, the reconstruction plate could not be fixed to the residual condylar region, necessitating disarticulation excision of the mandible. Moreover, this small condylar remnant may behave as a source of infection, postoperatively. With the advancement of new technology, three-dimensional virtual planning employing a stereolithographic model and navigation system can help design osteotomies and prevent injury to essential structures close to the tumor.[24] To address the controversy of immediate and delayed reconstruction, a marrow-frozen section was done and an Anterolateral thigh flap was harvested to cover the defect and reconstruction plate. This case report advocates immediate reconstruction provided radical surgical excision is planned and the intraoperative frozen section

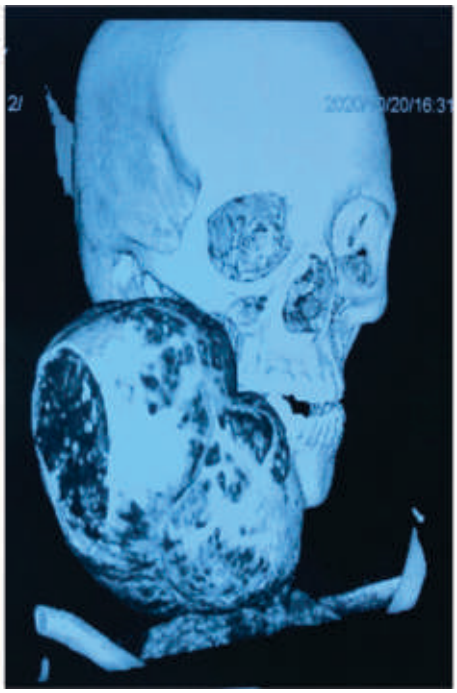
facility is available. The standard treatment for reconstruction of mandibular defects is a free vascularized bone flap e.g free fibula flap however due to venous insufficiency in both legs of the patient multidisciplinary team proposed an alternative option i.e Anterolateral thigh flap. The shortcoming of this flap is, it does not provide a bony template for dental implant placement and TMJ prosthesis fixation. But it is a good alternative in terms of coverage when a free fibula flap reconstruction is not possible. Limited follow-up guidelines are available, a close follow-up is however advised for PJOF, to detect recurrence.

CONCLUSIONS

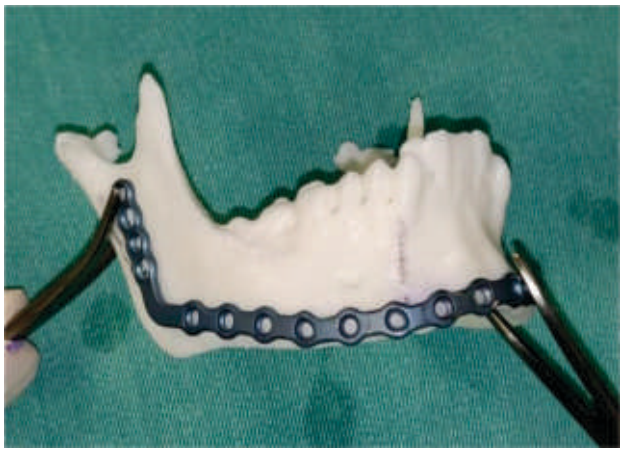
Psammomatoid juvenile Ossifying fibroma is a clinically challenging fibro-osseous lesion that warrants a definitive diagnosis. Detailed history, careful clinical examination, and contemporary imaging techniques (3D CT scan) aided by experienced histopathological reporting help in early diagnosis of the PJOF and ruling out potential differential diagnosis. A multidisciplinary team approach improves the quality of treatment. Recent advances include genetic testing for early diag-nosis of aggressive lesions. Management outcome largely depends on the location and duration of the disease. However, MDM2 and RASAL1 gene expre-ssion and the presence of aneurysmal bone cystic lesion's body are associated with a high recurrence rate and aggressive nature. Although, there is a debate, about whether to opt out conservative or aggressive approach. This report advocates an aggressive approach including radical excision and 0.5 cm safe margins. Modifying the incisions and dissection approaches is key to safe surgery. Stereolithographic models are of extreme help in planning the surgery and pre-bending of implants which will ultimately yield higher esthetic results. Disarticulation resection, an aggressive approach, is a key decision that should always be weighed in terms of patient benefits. Total joint replacement and free flap osteo-myo-cutaneous reconstruction can improve the patient's quality of life through comparatively better joint functions and dental rehabilitation by dental implants. There are two schools of thought regarding the sequence of reconstruction. This study supports immediate reconstruction provided the marrow frozen section is performed preoperatively. A free fibula flap is a reconstruction choice but if contraindicated antero-lateral thigh flap is an optimal alternative.



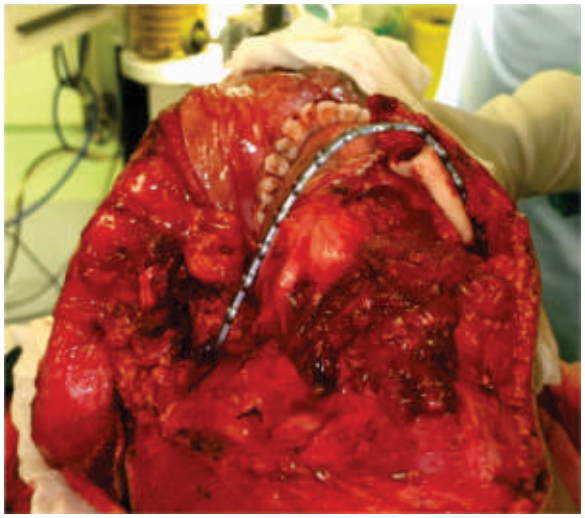
A. *Pre operative frontal facial profile*



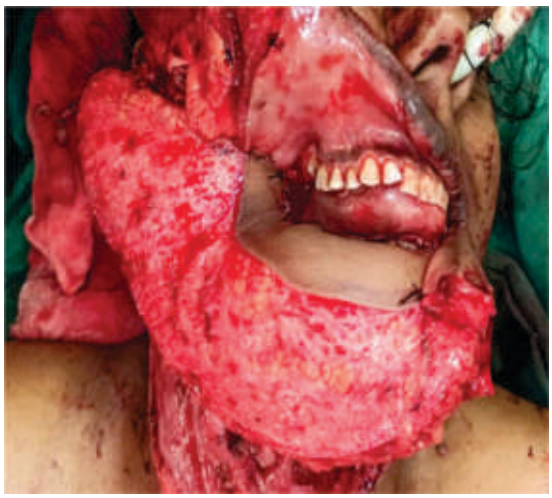
B. *Pre –OP CT scan face with 3D reconstruction*



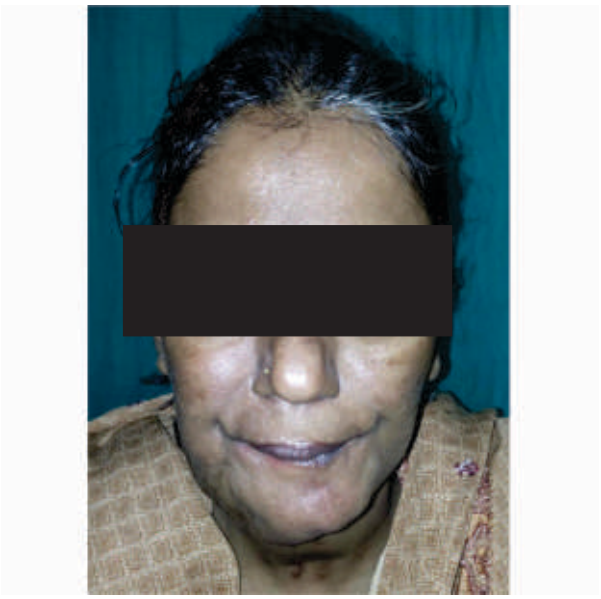
C. *VIRTUAL SURGICAL PLANNING ASSISTED 3D printed STL model for pre-bending of titanium reconstruction plate*



D. *Per-Op fixation of reconstruction plate*



E. *Per OP soft tissue reconstruction with Free Anteri-lateral thigh Flap*



F. *1 year post op frontal facial profile*

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