



Case Report

Extensive titanium mesh invasive cranial fibrous dysplasia

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ABSTRACT

Background: Craniofacial fibrous dysplasia (FD) is a benign lesion. It presents as bony swelling. Even after complete excision, it has a tendency to recur due to some residual lesion in normal bone. Recurrence at same site is common, but it recurs in bone. We are reporting a rare case of recurrent FD engulfing titanium mesh.

Case Description: A 22-year-old girl, who underwent frontal FD excision and reconstruction using titanium mesh surgery 2 years back, came with complaint of progressive bony swelling at same site for 1 year. CT head confirmed bony lesion involving mesh, frontal air sinus. She underwent complete excision of lesion and cranioplasty using bony cement. Biopsy confirmed recurrence of FD and invasion of titanium mesh.

Conclusion: Recurrence of FD, involving cranioplasty titanium mesh, is extremely rare. It suggests local invasiveness of lesion. Recurrence can be prevented by excision of lesion with free bony margins.

Keywords: Craniofacial, Fibrous dysplasia, Titanium mesh

INTRODUCTION

Skull fibrous dysplasia (FD) is a rare, slow-growing tumor, which mostly arises from frontal region.^[5] It is either monostotic, involving single bone, or polyostotic, involving more than 1 bone. Craniofacial FD management is mostly surgical. Recurrence, after surgery, is around 25–80%.^[1] It usually occurs at same site and involves bone. There is single case report of recurrent FD, which engulfs titanium mesh and few case reports about titanium plate invasion.^[4,6] We are reporting second such case of extensive titanium mesh cage invasion and its management.

CASE REPORT

A 23 years girl came with complaint of progressively painful swelling over forehead, 2 years ago. CT head confirmed FD of frontal bone and sinuses [Figure 1a and b] and recurrence of lesion almost 1 year after first surgery [Figure 1c-e]. She underwent excision of bony lesion and titanium mesh cranioplasty [Figure 2a-d]. After 1 year, she developed progressive hard swelling at same site [Figure 3a]. CT head confirmed engulfing titanium mesh and arising from frontal bone and sinus. [Figure 1c-d]. Her growth hormone and IGF level were normal. There was no family history. After informed consent, she underwent second surgery.

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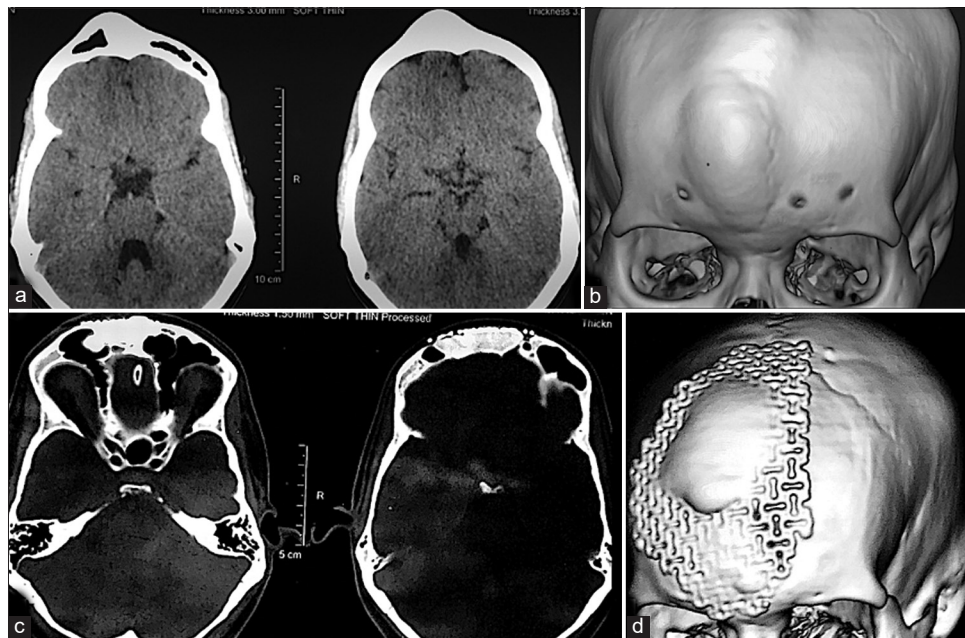


Figure 1: (a) CT head (2017), axial cut showing fibrous dysplasia arising from frontal bone, (b) 3D CT head (2017) showing fibrous dysplasia appearance on forehead, (c) CT head (2020) showing engulfment of titanium mesh, (d) 3D reconstruction depicting status of mesh and recurrent lesion.

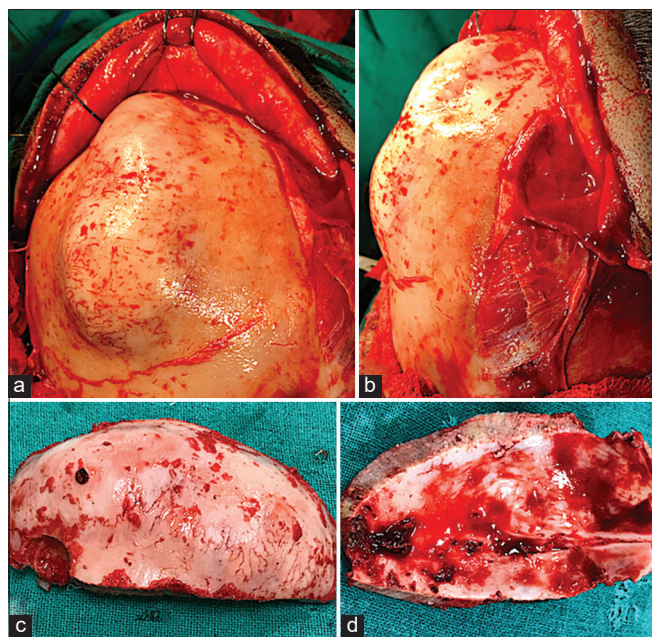


Figure 2: (a) Intraoperative image of fibrous dysplasia superior view, first surgery, (b) lateral view, (c) lesion resected with free bony margin, except frontal sinus area, anterior aspect, (d) posterior aspect.

Procedure

Bicoronal incision was given. The subcutaneous tissue was adhered to bony growth. It was sharply dissected free. The titanium mesh was completely engulfed by recurrent lesion

[Figure 3b and c]. Wide bony margins were resected to remove lesion *en bloc* with free bony margins [Figure 3d]. It was tightly adhered to dura, especially central part of mesh. The frontal sinuses were completely excised and exteriorized. Reconstruction was done with autologous bone and bone cement. Satisfactory contour was given [Figure 3e]. The patient was discharged after 10 days.

Follow-up

Follow-up after 3 months showed satisfactory result. Our plan is to repeat CT head after 1 year.

DISCUSSION

Craniofacial FD presents with cosmetic disfigurement, pain, vision loss, hearing impairment, and incidentally. It is an uncommon benign lesion, difficult to manage, when involves craniofacial region.^[1,5,6]

CT head with 3D reconstruction is investigation of choice.^[2] FD can be ground glass (most common appearance), sclerotic, or cystic on CT. CT image gives estimation of bony involvement, free bony margins, extent of cosmetic disfigurement, and spread into deeper or difficult location, for example, skull base or sinuses. MRI is indicated to find relation with cranial nerve or skull base.

Wu *et al.* reported a case of recurrent craniofacial FD engulfing titanium mesh.^[6] Similar to our case, it was involving frontal bone. Due to orbital involvement, some

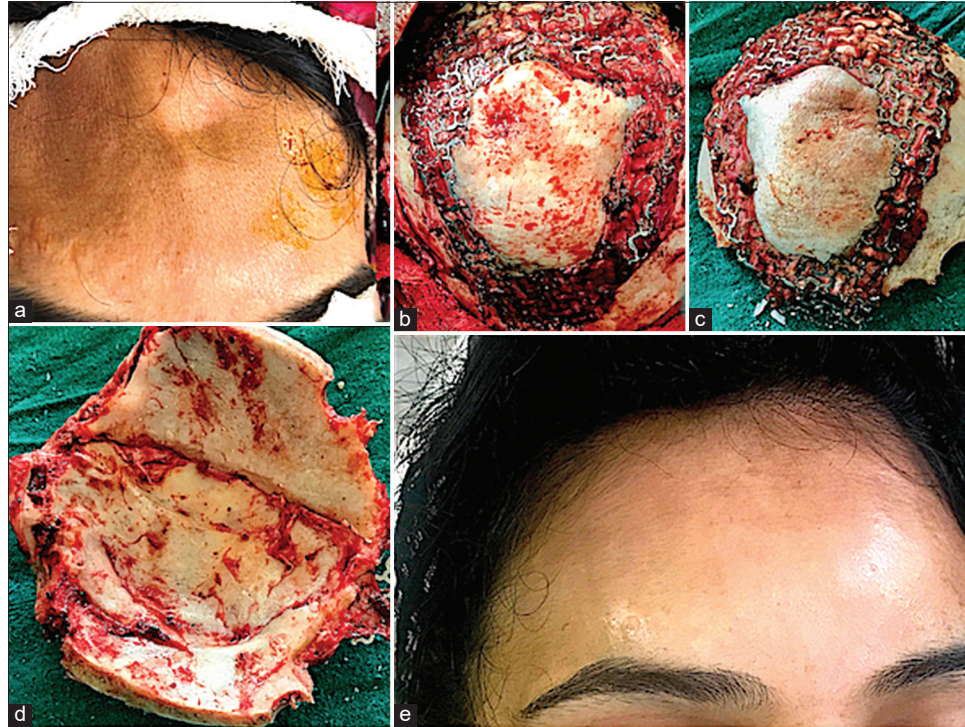


Figure 3: (a) Forehead shape before surgery, (b) intraoperative image showing recurrent fibrous dysplasia involving mesh, (c) resected lesion with mesh, anterior aspect, (d) posterior aspect, (e) forehead shape after surgery.

lesion was left in first surgery, which caused recurrence. According to authors, surgery can accelerate growth of residual lesion. In our case, residue was left in frontal sinus, which caused recurrence. Recurrence rate is around 71% after limited resection, around 15% after radical excision. Despite benign lesion, the recurrence rate is quite high. We also believe that surgery can accelerate growth of residual lesion. The level of hormones was found to be normal in our patient. It was done to find out any link between invasiveness of lesion and hormone.

The surgical management varies according to patient.^[1-4,6] In prepubertal age, wait is advised till puberty; unless there is functional loss. After puberty, FD growth slows down and excision gives better results. In asymptomatic cases, close observation is recommended. Single-stage radical excision and reconstruction with titanium mesh is ideal management for symptomatic FD. However, radical excision is limited by involvement of neurovascular structures and eloquent area.

There is no recommendation for reconstruction of bone after second recurrence. We chose bone cement instead of mesh to provide handmade cover for radically excised bone. Long-term result is awaited to confirm our treatment choice.

At present, no medical therapy has been found to be effective. However, it is important that any underlying endocrine

abnormalities be treated before surgical intervention.^[1] In McCune Albright syndrome and pediatric population, growth hormone excess is found to be cause of extensive FD.^[1,4]

CONCLUSION

Despite its benign nature, craniofacial FD has a tendency to recur after surgery. Frontal FD recurrence occurs due to missed lesion at deeper or difficult locations, for example, in sinus area. Recurrent lesion, with engulfed mesh, is extremely rare and difficult to excise. For best outcome, radical excision with free bony margins must be done.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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