ScientificScholar®
Knowledge is power
Publisher of Scientific Journals

www.surgicalneurologyint.com

Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, Clinical Professor of Neurological Surgery, School of Medicine, State U. of NY at Stony Brook.

SNI: General Neurosurgery

Editor
Eric Nussbaum, MD
National Brain Aneurysm and Tumor Center, Twin Cities, MN, USA

SNI
Open Access

Case Report

Calvarial hyperpigmentation

Devon van Eck[1], Blessing Ndlovu[1], Mohammed Ouwais Abdul Sattar[1], Denver Naicker[1], Morena Nthuse Mpanza[1], John Richard Bwire Ouma[2], Christos Profyris[1]

[1]Department of Neurosurgery, Helen Joseph Hospital, [2]Department of Neurosurgery, University of Witwatersrand, Johannesburg, South Africa.

E-mail: *Devon van Eck - Devonvaneck@gmail.com; Blessing Ndlovu - drbndlovu@gmail.com; Mohammed Ouwais Abdul Sattar - ouwais@live.co.za; Denver Naicker - denver.naicker@wits.ac.za; Morena Nthuse Mpanza - n2se@yahoo.com; John Richard Bwire Ouma - john.ouma@wits.ac.za; Christos Profyris - cp@drprofyris.com

***Corresponding author:**
Devon van Eck,
Department of Neurosurgery,
Helen Joseph Hospital,
Johannesburg, South Africa.

Devonvaneck@gmail.com

Received : 10 June 2023
Accepted : 31 July 2023
Published : 11 August 2023

DOI
10.25259/SNI_493_2023

Quick Response Code:

ABSTRACT

Background: Osseous hyperpigmentation of the calvarium is an extremely rare finding with only few reported cases in literature.

Case Description: The case is of a 59-year-old Caucasian male who presented with an acute history of generalized tonic clonic seizures and progressive weakness of the right upper limb. He had a background history of a malignant melanoma which had been resected from his left external acoustic meatus 4 weeks prior. Neuroimaging of the brain showed an intra-axial space-occupying lesion in his left parietal lobe with no associated osseous changes. A left mini parietal craniotomy was performed which revealed black discoloration of the parietal bone. The lesion was successfully resected and the bone flap was secured back in place. The patient was discharged on the 4th day postoperatively with no complications. The unusual finding of black discoloration of the calvarium was found to be secondary to adolescent tetracycline use.

Conclusion: Calvarial hyperpigmentation is a phenomenon encountered incidentally and will often come as surprise for surgeons. Once encountered, thorough history taking and examination should be done to investigate the cause.

Keywords: Black bone disease, Craniofacial, Osseous hyperpigmentation, Skull

INTRODUCTION

Calvarial osseous hyperpigmentation is a rare finding as evidenced by the sparse literature. To date, only five cases have been reported and published.

The most common cause of osseous hyperpigmentation is prolonged exposure to tetracycline during bone growth.[1] However, there are other sinister etiologies which should be excluded such as malignancy, infection, metabolic conditions, and various medications such as anti-leprosy or chemotherapeutic drugs and finally metallosis.[3]

CASE REPORT

A 59-year-old Caucasian male was referred to our unit after suffering from a 1 month history of progressive right upper limb weakness which was more prominent in the distal muscle groups. This was further complicated by an acute presentation of generalized tonic clonic seizures. He had a background history of resection of a malignant melanoma from his left external auditory meatus 1 month prior.

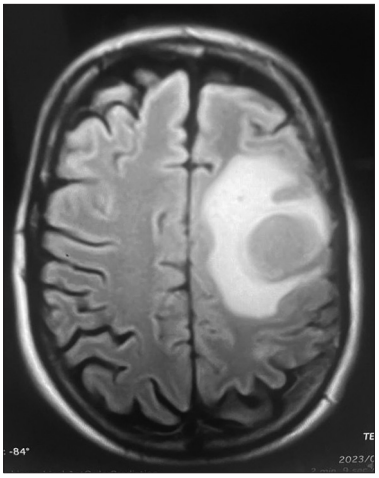

Figure 1: Axial T2-weighted fluid attenuated inversion recovery magnetic resonance imaging showing a left frontal intra-axial lesion with surrounding vasogenic edema.

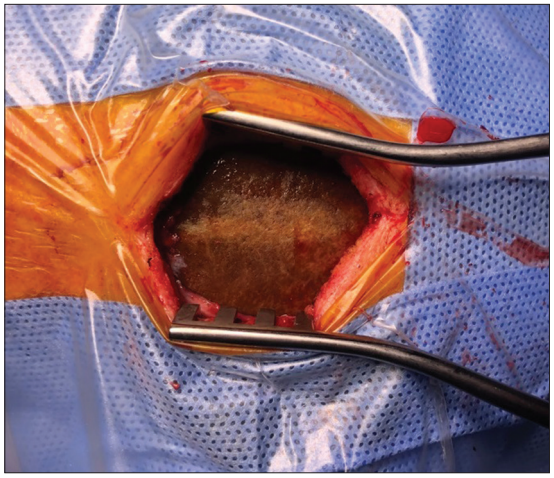

Figure 2: Intraoperative scalp dissection showing distinct discoloration of the parietal bone.

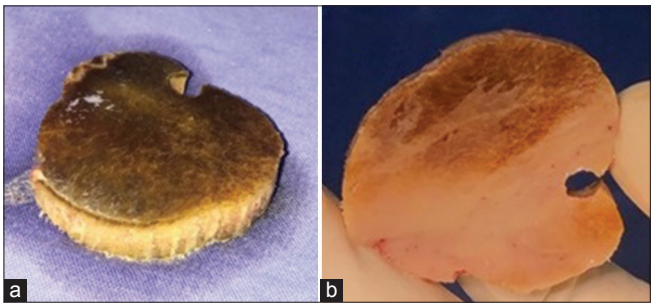

Figure 3: Bone flap showing discoloration of the outer table, parts of the diploe (a) as well as the inner table (b).

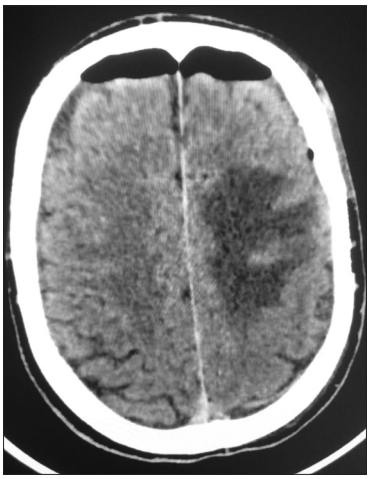

Figure 4: Postoperative axial contrasted computerized tomography of the brain showing gross total resection of metastatic malignant melanoma of the frontal lobe.

On computerized tomography (CT) and magnetic resonance imaging of the brain [Figure 1] a 3 cm well circumscribed space-occupying lesion with associated vasogenic edema were seen in the left frontal lobe. No osseous changes of the calvarium and skull base were noted on imaging. The clinical history and radiologic features were highly suspicious for metastatic melanoma. The patient was started on sodium valproate and dexamethasone before operative resection of the lesion.

Intraoperatively, a left parietal mini craniotomy was performed with assistance of neuro-navigation. Scalp dissection down to the bone revealed black discoloration of the parietal bone as depicted in Figure 2. The craniotomy bone flap was removed and on inspection showed hyperpigmentation of both the outer and inner tables and part of the diploe [Figure 3] without involvement of the underlying dura. These findings are unique to our case as all previously reported cases have described outer cortex involvement only. The bone quality and consistency appeared to be unaffected. Gross total resection of the brain lesion was then achieved.

There were no intraoperative nor immediate postoperative complications. The patient's focal neurological deficits improved with physiotherapy and no further seizures were noted during the admission. Further questioning and systemic examination showed no signs of hyperpigmentation elsewhere on the body, but of note, the patient admitted to a 3-year history of tetracycline use for adolescent acne vulgaris which we deduced to be the most probable cause of his calvarial hyperpigmentation. Immediate postoperative CT scans of the brain showed complete resection of the lesion [Figure 4] with histology confirming the diagnosis of metastatic malignant melanoma. The patient was then referred to oncology for further management. We continued to follow him up closely in our outpatient department and at 2 months postoperatively

he underwent a positron emission tomography scan which revealed no calvarial or intracranial hotspots.

DISCUSSION

Osseous hyperpigmentation of the calvarium is a very sparsely reported phenomenon. To best of our knowledge, only five cases have been reported in literature. Four cases described involvement of the frontal bone and only a single case affected the temporal bone. There seems to be a male preponderance in the reported cases with a male to female ratio of 4:1. All the previous cases reported no changes in bone quality or consistency and of the cases where craniotomy was performed, only the outer cortex was noted to be affected. In our case; however, we found hyperpigmentation of both the inner and outer cortices as well as the diploe.

There are several potential causes of bone hyperpigmentation such as malignancy, infection, metabolic conditions, chemotherapeutic drugs and metallosis. With regard to osseous hyperpigmentation of the calvarium, all the cases report prolonged exposure to tetracycline or minocycline as the cause. Bony hyperpigmentation secondary to tetracycline exposure is the most commonly stated source and is seen with cumulative doses of 100 g or more. The risk also increases with prolonged exposure with an additional 10% per year.[2]

Histologically and macroscopically no changes in bone quality or consistency have been noted, with *in vitro* studies showing increased bone density following 8 weeks of minocycline use.[5] The proposed mechanism of tetracycline induced osseous hyperpigmentation is due to the chelation of tetracycline molecules to calcium which are then deposited into mineralizing tissues during bone growth. These tetracycline calcium complexes then undergo oxidization and become discolored due to deposition of endogenous degradation products.[4]

There are no reported cases of radiographic changes in osseous hyperpigmentation.[2] This along with the above mechanism further supports the importance of detailed history taking which may be aided with careful examination of the teeth, maxillary and mandible mucosa to diagnose this condition preoperatively or retrospectively.

CONCLUSION

Calvarial hyperpigmentation is a rare incidental finding which can be encountered intraoperatively. Although most commonly caused by adolescent tetracycline use, it is important to rule out other sinister causes.

Declaration of patient consent

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

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Chubb DW, Finkemeyer JP, Bush KL. Etiology of craniofacial bone hyperpigmentation and an algorithm for addressing intraoperative discovery. Plast Reconstr Surg Glob Open 2020;8:e3061.
2. Eisen D, Hakim MD. Minocycline-induced pigmentation. Incidence, prevention and management. Drug Saf 1998;18:431-40.
3. Farahnik B, Zaghi S, Hendizadeh L, Gopen Q. Rusty green stained temporal bone associated with exposure to tetracycline: An unusual presentation of black bone disease. J Laryngol Otol 2015;129:276-8.
4. Sánchez AR, Rogers RS 3rd, Sheridan PJ. Tetracycline and other tetracycline-derivative staining of the teeth and oral cavity. Int J Dermatol 2004;43:709-15.
5. Williams S, Wakisaka A, Zeng QQ, Barnes J, Martin G, Wechter WJ, et al. Minocycline prevents the decrease in bone mineral density and trabecular bone in ovariectomized aged rats. Bone 1996;19:637-44.

How to cite this article: van Eck D, Ndlovu B, Abdul Sattar MO, Naicker D, Mpanza MN, Ouma JR, *et al*. Calvarial hyperpigmentation. Surg Neurol Int 2023;14:289.