



Total Resolution of Large Scalp Swelling due to Calvarial Tuberculosis with Medical Management Only: Case Report and Review of the Literature

Kalvariyal Tüberküloz Nedeniyle Geniş Saçlı Deri Şişmesinin Sadece Tıbbi Yaklaşımla Tamamen Geçmesi: Olgu Raporu ve Literatür Derlemesi

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ABSTRACT

Calvarial tuberculosis is rare. It presents with single or multiple discharging sinus or a localized focal scalp swelling with overlying skin discoloration. Usually subgaleal collection is associated with evolution of sinus formation in different stages. However, extensive scalp swelling spreading across bifrontal area and overlying healthy scalp is not reported. Pathologically it causes significant destruction of calvarium including erosion of both the tables with concurrent significant epidural granulation tissue deposits as dura acts as barrier to trans-dural spread. Concurrent surgical management and antitubercular medication is the standard treatment modality.

Authors report a unique presentation of calvarial tuberculosis in 17-year- female presented with complaint of large painless scalp swelling extensively spreading over both frontal area from supraorbital margin to coronal suture without discoloration, puckering of overlying scalp or sinus formation, and neuroimaging showed absence of extradural granulation tissue deposits with very subtle bony architecture changes of adjacent calvarium. The swelling completely subsided with medical therapy alone. To the best of knowledge of authors such case of calvarial tuberculosis having large scalp swelling in western literature has not been reported. Clinical presentation, imaging, management and pertinent literature are reviewed.

KEYWORDS: Calvarial tuberculosis, Tubercular large subgaleal collection, Management, Tubercular osteomyelitis, Extrapulmonary skeletal tuberculosis

ÖZ

Kalvariyal tüberküloz nadirdir. Ciltte renk değişikliğiyle birlikte boşalan tek ya da çok sayıda sinüs veya lokalize fokal saçlı deri şişmesiyle ortaya çıkar. Genellikle subgaleal kolleksiyon farklı evrelerde sinüs oluşumunun gelişimiyle ilişkilidir. Ancak bifrontal bölge ve üzerindeki sağlıklı saçlı deri üzerinde yayılan geniş saçlı deri şişmesi daha önce bildirilmemiştir. Patolojik olarak kalvariyumda önemli harabiyete yol açar ve her iki tablada erozyonla birlikte transdural yayılmaya bir bariyer etkisi gösteren eş zamanlı önemli epidural granülasyon dokusu kalıntıları mevcuttur. Standart tedavi modalitesi cerrahi tedavi ve antitüberküloz ilaçların birlikte uygulanmasıdır.

Yazarlar saçlı deride herhangi bir renk değişikliği, çukurlaşma veya sinüs oluşumu olmadan supraorbital kenardan koronal suture kadar her iki frontal bölge üzerine geniş olarak yayılan ve nörogörüntülemeye ektradural granülasyon dokusu kalıntıları bulunmayıp komşu kalvariyumda çok hafif kemik mimarisi değişiklikleri saptanan 17 yaşında bir kadın hastayı sunmaktadır. Şişme sadece tıbbi tedaviyle tamamen geçmiştir. Bildiğimiz kadarıyla batı literatüründe böyle geniş saçlı deri şişmesiyle bir kalvariyal tüberküloz olgusu bildirilmemiştir. Klinik prezentasyon, görüntüleme, takip ve ilgili literatür gözden geçirilmiştir.

ANAHTAR SÖZCÜKLER: Kalvariyal tüberküloz, Tüberküler büyük subgaleal sıvı toplanması, Yönetim, Tüberküler osteomyelit, Ekstrapulmoner iskelet tüberkülozu

ABBREVIATIONS: CT: computed tomography, **ESR:** erythrocyte sedimentation rate, **HU:** Hounsfield unit, **ATT:** anti-tuberculosis therapy

INTRODUCTION

Tuberculosis is endemic in developing countries due to common prevalence of malnutrition, poor sanitation, and lack of hygiene awareness, drug defaulter, and drug resistance. Recently there is also surge in cases of tuberculosis in developed countries, due to association and increasing prevalence of acquired immunodeficiency disease (4). Calvarial tuberculosis is a rare occurrence with only few case series has been published (3, 6, 11, 13). It usually presents with small painless swelling, with chronic discharging sinus. Pathologically it is characterized by varying degree of erosion or destruction of inner and outer table of skull and may be associated with epidural collection on inner aspect or subgaleal collection on outer aspect. We report a unique case of calvarial tuberculosis in a 17-year-old female presented with large bifronto-parietal painless scalp swelling due to subgaleal collection with almost normal skull bony architecture on initial CT scan imaging. There was minimal erosion of outer table on repeat CT scan which led to dilemma in management. It accumulated repeatedly after tapping. However, on initiation of antituberculous treatment it responded well and did not require surgical intervention.

CASE REPORT

A 17-year-old girl residing in rural area was referred to our neurosurgical services by a local practitioner, with an insidious onset large bifronto-parietal painless swelling. There was no associated fever, evening rise of temperature, weight loss. She could not recall any trauma to head in recent six months. She had no other significant medical illness in the past. Her mother had pulmonary tuberculosis for which she received a full course of antituberculous treatment 1 year back. Her main concern was cosmetic disfigurement (Figure 1A-C) for which aspiration was done by local physician which revealed haemorrhagic aspirate. Cyto-pathological examination of aspirate did not reveal any malignant cells and showed only chronic inflammatory cells, Ziehl-Nielsen staining was

negative. Clinical examination revealed large soft, fluctuant swelling extending from supraorbital margin and extending across bifrontal region to few centimetres posterior to the coronal suture. Overlying skin was healthy and no discharging sinus was noted. The rest of the physical and neurological examination was unremarkable. Haematological examination revealed mild leucocytosis with lymphocytosis, ESR was raised. Mantoux test was positive.

The initial CT scan of head revealed a large heterogeneous soft tissue collection in subgaleal space on topogram, size measuring about 10x7x2 cm with scattered hyperdense foci (HU-40-60) (Figure 1A-C). Bone window revealed normal diploe with intact inner and outer table without epidural collection

So a decision to start antituberculous therapy (ATT) was taken, and after 10 days it showed gradual diminution in size and disappeared completely after six weeks of treatment. Repeat computed tomography after six weeks showed small subgaleal collection without epidural collection and bone window revealed normal diploe with subtle erosion of outer cortex, with intact inner table (Figure 2A-B). She responded well to conservative medical antituberculous therapy. CT scan after 8 months of therapy revealed complete resolution of subgaleal collection and bony changes (Figure 3).

DISCUSSION

Tuberculosis is endemic in developing countries and there has been a surge in developed countries as well due to HIV infection (4). It is caused by *Mycobacterium tuberculosis*. Calvarial tuberculosis is a rare site of extra-pulmonary skeletal tuberculosis (2). The lung is the usual primary site. Occurrence of systemic including skeletal tuberculosis is usually secondary and the haematogenous route is the commonest mode of spread (1, 9). *Mycobacteria* get trapped in cancellous part of calvarial bone reaching from pulmonary, renal or rarely as a part of miliary tuberculosis (15).

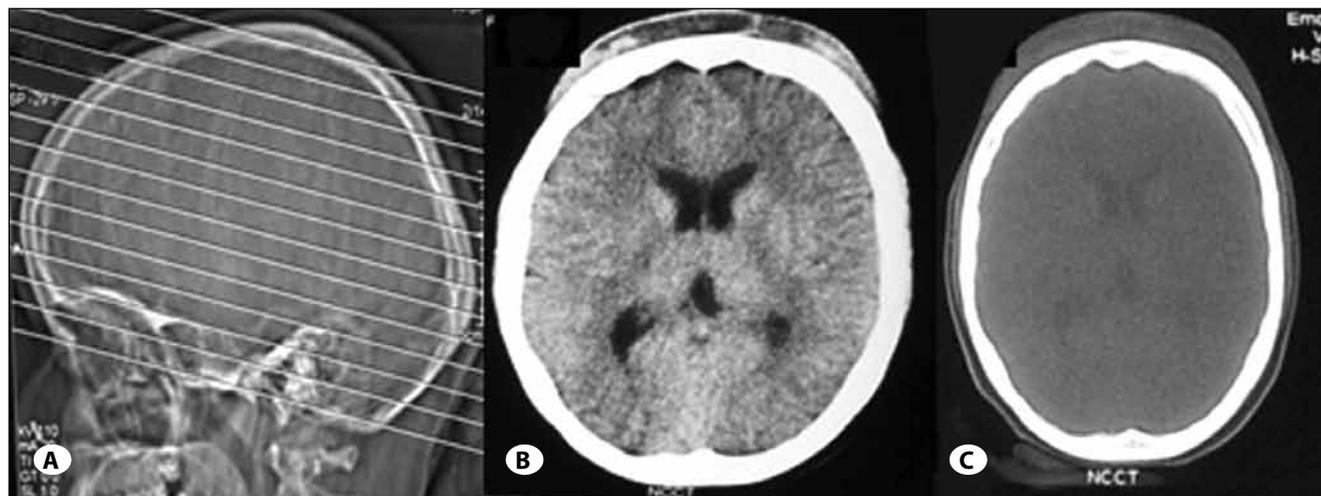


Figure 1: Initial NCCT head showing: **A)** topogram revealing large bifronto-parietal scalp swelling, **B)** axial section showing scalp soft tissue swelling with normal bone and no intra-parenchymatous lesion, **C)** axial bone window showing normal bony architecture.

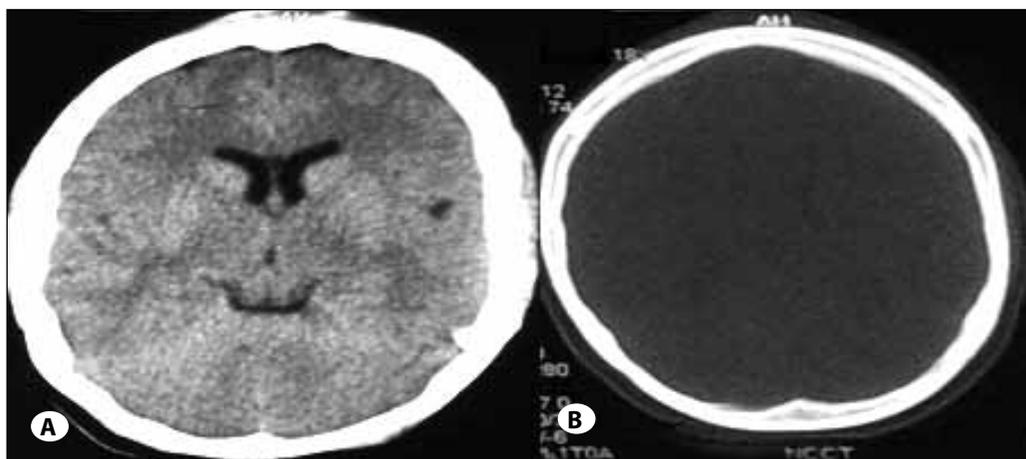


Figure 2A, B: NCCT head after 6 weeks following initiation of ATT, showing reduction in size of swelling, with subtle evidence of bony erosion.

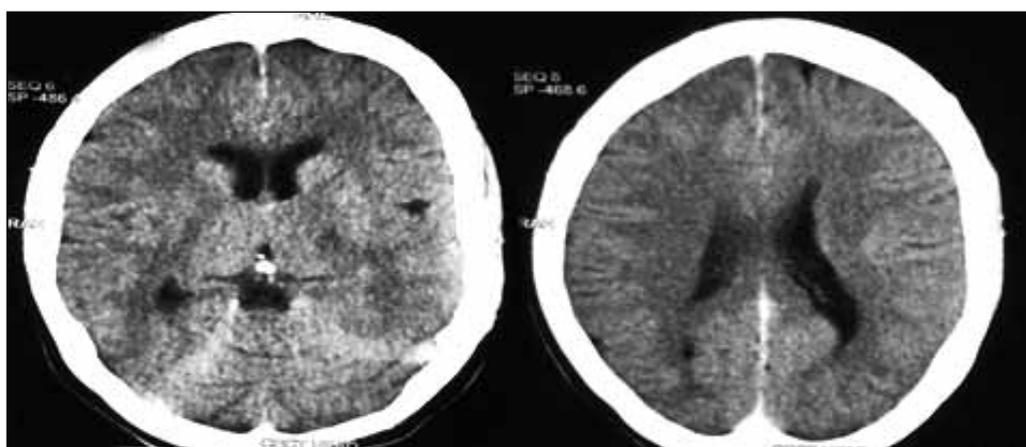


Figure 3: CECT head after 8 months of ATT therapy showing complete resolution of scalp swelling.

This leads to accumulation of lymphocytes, plasma cells, focal polymorphonuclear cells, Langerhans giant cells with caseation necrosis. Further expansion of the lesion causes erosion of inner or outer table leading to destruction and collection in epidural or subgaleal space (5). Subgaleal collection may rupture spontaneously to form chronic discharging sinus and may have more than one sinus tract. Subgaleal collection, if small, may cause adherence to overlying skin with discolouration (15).

It commonly involves frontal or parietal bone (3, 9, 13). Sutures usually do not pose any resistance so it can easily spread across suture lines and involve adjacent bone. However, intact dura is resistant, so subdural collection including meningitis or cerebral abscess is usually not seen (9, 10). That may be the reason of preserved neurological status and absent meningeal signs.

Three types of calvarial tuberculosis have been described on conventional radiography depending on the nature of calvarial destruction. Volkman (16) described perforating type calvarial tuberculosis characterized by small circumscribed punched out bony defect with granulation tissue covering both inner and outer aspect with minimal tendency of osteitis surrounding the defect. Koenig (7) used the term "diffuse tuberculosis of the cranium" for lesions causing widespread

destruction of the inner table of the skull. When these lesions are associated with extradural granulation tissue, they have been redefined as "spreading-type" lesions (7). Mohanty et al., have reported the third and least common type, the circumscribed and sclerotic type in which there is marked thickening of the bone because of lack of blood supply to the diseased bone (10).

The authors have reported management of 21 cases of calvarial tuberculosis all of them were surgically managed with debridement and removal of osteomyelitic bone and granulation tissue (13). Three patients required craniotomy for associated extensive extradural component. All patients were given ATT for eighteen months (13).

We postulate reactive effusion in subgaleal space with absent epidural component and almost preservation of bony architecture. The paucity of collection in epidural space may be due to dural resistance and or pressure gradient across epidural and subgaleal space. Furthermore it may be due to low virulence of organism, relative preserved immune status or good nutritional status, as our patient was in good health.

Management of calvarial tuberculosis is challenging in the absence of consensus. Surgery has been advocated as the main modality and includes scalp flap raising, removal

of extradural collection, scrapping of eroded bones, and excision the of sinus tract, with primary closure. Raut et al. (14) reported good outcome of all patients who had radiological evidence of epidural collection. Surgery led to good outcome, furthermore only 1 out of 11 cases was managed without surgery.

Many authors have advocated only medical treatment and have also reported good outcome following medical therapy (8, 12). Ramdurg et al. recommend minimum of eighteen months of antituberculous chemotherapy in all cases while Raut et al. suggested only nine months to one year (13, 14). Medical treatment may form initial part of treatment in those cases detected early, having no epidural collection or destruction of bone. However, a possibility of surgical option should also be kept as and when treatment fails, or development of multidrug resistance, poor compliance of drug, large epidural or progressive growing collection, formation of fresh sinus tract or amount of discharge output not reducing or cosmetic disfigurement (6, 8). Our case was managed without surgical intervention. However, very few selected cases may be given trial of medical treatment and keeping surgical option as stand by procedure as and when required or warranted (3). Surgical intervention may be required especially where diagnosis is in doubt or failure to respond to medical treatment or purely cosmetic deformity.

CONCLUSION

In secretory or exudative type of calvarial tuberculosis, conservative antituberculous drug treatment may be given as initial line of treatment with surgery kept as second line treatment as and when required or warranted.

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