



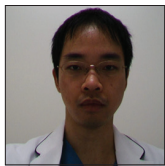
## Case Report

# Frontotemporal dermoid cyst with incomplete dermal sinus tract in an adult: A case report

Sukwoo Hong<sup>1</sup>, Keisuke Maruyama<sup>1</sup>, Ryo Hatanaka<sup>1</sup>, Akio Noguchi<sup>1</sup>, Hiroaki Shimoyamada<sup>2</sup>, Motoo Nagane<sup>1</sup>, Yoshiaki Shiokawa<sup>1</sup>

Departments of <sup>1</sup>Neurosurgery, <sup>2</sup>Pathology, Kyorin University Hospital, Mitaka, Tokyo, Japan.

E-mail: \*Sukwoo Hong - honsokhaisei6031@gmail.com; Keisuke Maruyama - kskmaru-tyk@umin.ac.jp; Ryo Hatanaka - r-htnka@hotmail.co.jp; Akio Noguchi - nogu-kyr@umin.ac.jp; Hiroaki Shimoyamada - hshimoya@ks.kyorin-u.ac.jp; Motoo Nagane - mnagane@ks.kyorin-u.ac.jp; Yoshiaki Shiokawa - shiokawa-umin@ks.kyorin-u.ac.jp



### \*Corresponding author:

Sukwoo Hong,  
Department of Neurosurgery,  
Kyorin University Hospital,  
Mitaka, Tokyo, Japan.

[honsokhaisei6031@gmail.com](mailto:honsokhaisei6031@gmail.com)

Received : 09 August 2020  
Accepted : 17 November 2020  
Published : 11 December 2020

DOI  
[10.25259/SNI\\_504\\_2020](https://doi.org/10.25259/SNI_504_2020)

Quick Response Code:



## ABSTRACT

**Background:** Non-midline supratentorial dermoid cyst with dermal sinus tract has been rarely reported especially in adults. We recently experienced a noteworthy patient with frontotemporal dermoid cyst with incomplete dermal sinus tract.

**Case Description:** A 43-year-old female presented with recurrent subcutaneous mass in the left superolateral orbital region. She had a history of active bronchial asthma, which precluded her from contrast-enhanced imaging studies. Plain imaging studies showed a subcutaneous mass which was continuous with an intrasylvian fissure mass by a tract in the sphenoid ridge and the lesser wing of the sphenoid bone. Frontotemporal craniotomy was performed to reset the mass and the tract. Intraoperative finding showed no intradural tumor components. Extradural component was carefully removed focusing attention on the frontal branch of the facial nerve. The pathology was consistent with dermoid cyst and dermal sinus tract. Postoperatively, she had mild facial palsy of the corrugator supercilii (House and Brackmann Grade II). She was discharged home with modified Rankin scale 1.

**Conclusion:** Dermoid cyst needs to be included in the differential diagnosis of adult-onset subcutaneous mass in the frontotemporal regions. After thorough imaging studies for the presence and extent of the sinus tract, the symptomatic lesion should be excised completely once and for all.

**Keywords:** Adult, Dermal sinus tract, Dermoid cyst, Frontotemporal, Pterion

## INTRODUCTION

Dermoid cyst is typically a congenital benign tumor considered to be derived from abnormal ectodermal fusion<sup>[9]</sup> during fetal development. It accounts for 0.04–0.7% of all intracranial tumors.<sup>[4,12,20]</sup> Intracranial dermoid cyst tends to occur in the midline<sup>[9]</sup> whereas scalp (extracranial) dermoid cyst may occur off-midline.<sup>[18]</sup> However, in the past literature, we have several rare reports on these intracranial non-midline dermoid cysts associated with dermal sinus tract, especially in children.<sup>[2-4,6]</sup> We recently experienced a similar case who was diagnosed in adult. This case reminds us that non-midline dermoid cyst with continuous sinus tract may go undiagnosed until adulthood. This type of dermoid cyst sinus tract complex has not been reported, to the best of our knowledge. We reviewed the past literature similar to our case to shed light on the frontotemporal dermoid cyst sinus tract complex with emphases on its characteristics and management strategy.

This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

©2020 Published by Scientific Scholar on behalf of Surgical Neurology International

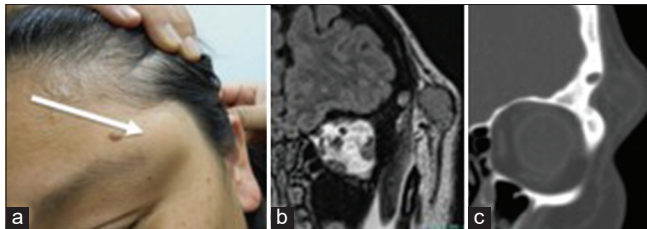
## CASE REPORT

### History and examination

A 43-year-old female presented with recurrent subcutaneous mass in the left superolateral orbital region [Figure 1a]. The mass has been present since she was 4 years old, and there have been no episodes of local infections. She had a history of partial resection of this mass at another department 2 years ago, where only subcutaneous component of the cyst was ligated and resected. Pathological diagnosis was dermoid cyst. In 2 years' follow-up, she developed a recurrent mass in the same region. Due to her active asthma and the history of adverse events that occurred after contrast MR imaging, contrast imaging studies were not performed. CT and MR imaging showed a subcutaneous mass with possible continuity to the Sylvian cistern by a tract in the sphenoid ridge and lesser wing of the sphenoid bone [Figure 1b and c]. As 2 years have passed since the initial surgery, we speculated the lesion had already penetrated the subarachnoid space through the dura based on the preoperative imaging. While she had no headache or showed any signs of neurological deficits, we decided to reset the mass and the tract to prevent further recurrence.

### Surgical intervention

She underwent frontotemporal craniotomy. When musculocutaneous flap was retracted, the cyst duct was inadvertently damaged, resulting in spill out of greasy and whitish yellow cyst content [Figure 2a]. The tumor was



**Figure 1:** Preoperative images of the recurrent dermoid cyst (a). Note the subcutaneous swelling in the superolateral orbital area (white arrow) Fluid attenuated inversion recovery MR imaging (coronal) showed a well-circumscribed extracranial tumor component of 22 mm in diameter (b). CT (coronal) showed a tract traversing through the sphenoid bone (c).



**Figure 2:** Intraoperative findings. Part of the tract was inadvertently damaged and greasy and whitish yellow tumor content spilled out (a). The bone around the tumor tract was drilled and the tract was dissected (b), which ended in pouch at the surface of the dura (c).

relatively large and fanning toward the skin, suggesting that dissection of the tumor through the temporal muscle and fascia could lead to damage to the frontal branch of the facial nerve.<sup>[16]</sup> We thus cut the cyst duct and followed the duct toward the bone [Figure 2b] and to the dura. The duct looked ending in a blind pouch at the surface of the dura [Figure 2c] near the lesser wing of the sphenoid bone. As the lumen was exposed, yellowish debris and hair were appreciated. Intradural space was then carefully inspected and showed no evidence of lesion continuity. Finally, the remaining extracranial mass was totally resected with careful inspection of the facial nerve using a nerve stimulator.

### Histopathological findings

The resected specimens showed a cystic lesion filled with keratinized debris and a ductal structure which was connected to the cyst. The cyst wall was lined by a stratified squamous epithelium similar to epidermis. It contained adnexa of skin, such as sebaceous glands and sweat glands [Figure 3]. As for the transitional segment from the cystic lesion and the dura, histiocytes infiltration and hyalinized dura were appreciated, with no dermoid components. Consequently, dermoid cyst and sinus tract were diagnosed.

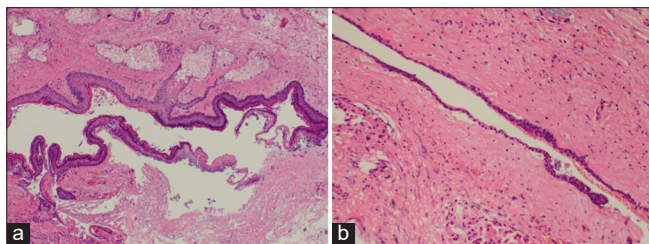
### Postoperative course

The patient developed mild facial palsy of the corrugator supercilii. House and Brackmann grade was 2. She was discharged home 8 days after operation with modified Rankin scale of 1. At 6 months after the second surgery, she was free of recurrence clinically and radiologically.

## DISCUSSION

A noteworthy case of subcutaneous dermoid cyst in the left superolateral orbital region accompanied by incomplete dermal sinus tract was described. Even though quite a few case reports have been made despite its rarity,<sup>[1-4,8,10-15,17,19-22]</sup> our case was unique in that it was diagnosed in adult and that the dermoid cyst was associated with incomplete sinus tract. Nineteen cases of frontotemporal dermoid cysts with sinus tract including ours are summarized in Table. The average age of diagnosis was 6 years. The lesions seemed to be more

common in females (five male patients and 14 female patients;  $P = 0.03$  by Chi-squared test) and more common on the right side (right in 13 cases, left in 5, unknown in 1;  $P = 0.06$  by Chi-squared test). The relapses occurred in five patients



**Figure 3:** A cystic lesion lined by an epidermal-like squamous epithelium contained several sebaceous glands, some of which continuous to the epithelium (a). A ductal structure, traversing through the sphenoid bone, was lined by a thin squamous epithelium with seldom keratinization (b). H&E: Hematoxylin and eosin; Original magnification  $\times 40$ .

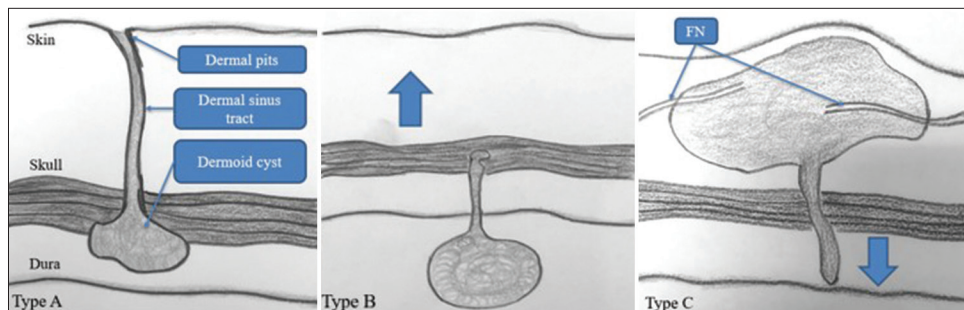
(26%) due to incomplete excision, but no recurrences were reported after complete excision with the median follow-up period of 8 months (range 1.5 months–8 years). The lesions were extradural in most cases. Dural transgression were reported in only two cases (10%), which was different from the midline lesions in the spine.<sup>[7]</sup>

Based on these previous reports, we classified the dermoid cyst sinus tract complex into three types (types A, B, and C) [Figure 4] by the location of the sinus tract relative to the dermoid cyst [Table 1]. Type A is characterized by the clinically overt dermal sinus tract ending in pouch with dermoid cyst in the intradiploic, extradural, or intradural areas. This type is diagnosed exclusively in the childhood due to recurrent local infections and discharge from the overt frontotemporal pits. Type B is characterized by the incomplete dermal sinus tract extending outward (toward the skin) from the intradural dermoid cyst. This type may be diagnosed in adults when they become large enough

**Table 1:** Past intracranial surgical reports on the frontotemporal dermoid cyst- sinus tract complex. All underwent complete excisions in the end and none recurred thereafter. Most of the cases were diagnosed in children and shared the same positional relationship of the cyst and the tract. The two unique adult cases were put in the lowest two rows for comparison.

Authors, years	Age/sex	Signs and symptoms	Side	Previous incomplete excisions	Type	Postoperative course
Neblett <i>et al.</i> , 1970 <sup>[14]</sup>	1/M	Local swelling	R	N	A	No recurrence at 3 year
Green, 1973 <sup>[8]</sup>	6/M	Local swelling	R	N	A	No recurrence at 9 months
Cullen, 1974 <sup>[6]</sup>	2/F	Discharging sinuses	R	N	A	
Hong, 1998 <sup>[10]</sup>	2/F	Discharging sinus	R	N	A	
Niederhagen <i>et al.</i> , 1998 <sup>[15]</sup>	1/M	Draining sinus	R	N	A	
Meyer <i>et al.</i> , 1999 <sup>[13]</sup>	2/M	Discharging sinus, orbital cellulitis	R	N	A	No recurrence at 8 years
Akita <i>et al.</i> , 2000 <sup>[11]</sup>	3/F	Discharging sinus, local infection	R	Y	A	No recurrence at 6 months
Parag <i>et al.</i> , 2001 <sup>[17]</sup>	1/F	Discharging sinus	L	N	A	
Lacey <i>et al.</i> , 2003 <sup>[11]</sup>	2/F	Discharging sinus, orbital cellulitis	L	Y	A	
	5/F	Discharging sinus	L	N	A	
	5/F	Discharging sinus, local swelling	R	Y	A	
Wells and Harris, 2004 <sup>[22]</sup>	0/M	Discharging sinus, orbital cellulitis	ND	N	A	No recurrence at 7 months
Scolozzi <i>et al.</i> , 2005 <sup>[19]</sup>	1/F	Dermal pits	R	N	A	No recurrence at 8 months
Mack and Ghatan, 2007 <sup>[12]</sup>	1/F	Discharging sinus	L	N	A	No recurrence at 1 year
Barnard <i>et al.</i> , 2012 <sup>[3]</sup>	3/F	Local infection, vomiting	R	Y	A	
Bliss <i>et al.</i> , 2016 <sup>[4]</sup>	3/F	Discharging sinus, orbital cellulitis	R	N	A	Transient paresis of FBFN No recurrence at 1.5 months
Barkley <i>et al.</i> , 2019 <sup>[2]</sup>	1/F	Discharging sinus, orbital cellulitis	R	N	A	No recurrence at 3 months
Shehadi <i>et al.</i> , 1999 <sup>[20]</sup>	31/F	Seizure	R	N	B	No recurrence at 1 year
Our case, 2020	43/F	Local swelling	L	Y	C	Transient paresis of FBFN No recurrence after 6 months

R: Right, L: Left, N: No, Y: Yes, ND: Not described in the abstract, FBFN: Frontal branch of the facial nerve



**Figure 4:** Dermoid cyst sinus tract complex is classified into three types based on the positional relationship of cyst and tract. In type A, the complete tract begins from the skin surface and the cyst is along the tract. Please note the relationship of the dermal pits, sinus tract and the cyst to the skin, skull, and the dura. Type B is the intradural dermoid cyst and the incomplete tract extending outward to the skin (arrow). Type C is the dermoid cyst and the incomplete sinus tract extending inward to the dura (arrow). In this type, the facial nerve (FN) branches may be around the cyst mass.

to cause mass effect.<sup>[20]</sup> Type C is also characterized by the incomplete dermal sinus tract extending inward (away from the skin) from the extracranial dermoid cyst. This type tends to be diagnosed in adults, although more thorough imaging would make it possible to diagnose in children if they presented with subcutaneous swelling. Based on our case (Type C), we believe, this type has a unique embryological mechanism different from the other types A and B, because the vector of the sinus tract is 180° different. It is unlikely that this lesion (Type C) was caused by the previous incomplete surgical resection, as the intracranial component and the sinus tract through the skull had already been present in the MR imaging before the index surgery.

### Management strategy

Reviewing the literature, in Type A, where recurrent skin infections leads to the diagnosis in childhood, surgical excision of the lesion is the treatment of choice.<sup>[2,10]</sup> Conservative or incomplete surgical resection almost always led to the relapses. Thus, when children presented with frontotemporal dermal pits and local inflammation, it is important to consider the possibility of a dermal sinus tract and associated dermoid cyst and to evaluate them with CT or MRI to achieve complete resection. In Type B, which should present with symptoms related to intracranial mass lesions such as headache, nausea, or seizure, a non-midline dermoid cyst should be included as a differential diagnosis of intradural mass lesions. In this type (B), since the tract is not obvious from the physical examination, it is important to evaluate radiologically for the presence of the sinus tract. Since incomplete surgical excision most likely leads to the relapse of the symptoms, complete surgical excision is warranted. In Type C, patients may be adults and present with local swelling near the temple. Dermoid cyst should be taken into the differentials and should be evaluated carefully with CT or MR imaging for the extent of the mass and the presence of the sinus tract. Since these lesions are benign, treatment

may be reserved until when they are symptomatic or for cosmetic reasons. Complete excision should be attempted to prevent relapse. In a series of 33 adults' dermoid cysts in the frontotemporal regions (none were associated with sinus tract), recommended preoperative evaluation using CT for masses larger than 2 cm.<sup>[5]</sup> MR imaging is useful if the cyst has associated sinus tract extending intracranially,<sup>[10,11]</sup> which was true in our case as well. Moreover, MR imaging with contrast medium may be helpful in equivocal cases like ours to evaluate the presence of intradural tumor components, which may omit the need to inspect the intradural space.

### CONCLUSION

Dermoid cyst needs to be included in the differential diagnosis of adult-onset subcutaneous mass in the frontotemporal regions. After thorough imaging studies for the presence and extent of the sinus tract, the symptomatic lesion should be excised completely once and for all.

### 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. Akita S, Hirano A, Fujii T. Recurrent, discharging congenital frontotemporal dermoid cyst. *Ann Plast Surg* 2000;44:465-6.
2. Barkley AS, Susarla SM, Lee A. Frontotemporal dermal sinus tract with 2 connected intradiploic dermoid cysts: A rare case

- and review of the literature. *World Neurosurg* 2019;127:350-3.
3. Barnard AR, Jones AP, Hodgkinson PD, Jenkins AJ. Beware frontotemporal dermoids-they may have intracranial extension: A case of a middle cranial fossa cyst. *J Plast Reconstr Aesthet Surg* 2012;65:e185-8.
  4. Bliss M, Grant G, Tittler E, Loven T, Yeom KW, Sidell D. Diagnosis and treatment of pediatric frontotemporal pits: Report of 2 cases. *J Neurosurg Pediatr* 2016;18:471-4.
  5. Chang JW, Yoon JS, Lee JH. The appropriate surgical approach to frontotemporal dermoid cysts in adult patients. *Ann Plast Surg* 2017;78:54-8.
  6. Cullen JE. Orbital diploic dermoids. *Br J Ophthalmol* 1974;58:105-6.
  7. Elton S, Oakes WJ. Dermal sinus tracts of the spine. *Neurosurg Focus* 2001;10:e4.
  8. Green MF. A lateral dermal sinus associated with an intradiploic cyst. *Br J Plast Surg* 1973;26:298-300.
  9. Greenberg MS. *Handbook of Neurosurgery*. 9<sup>th</sup> ed. Germany: Thieme; 2016.
  10. Hong SW. Deep frontotemporal dermoid cyst presenting as a discharging sinus: A case report and review of literature. *Br J Plast Surg* 1998;51:255-7.
  11. Lacey M, Gear AJ, Lee A. Temporal dermoids: Three cases and a modified treatment algorithm. *Ann Plast Surg* 2003;51:103-9.
  12. Mack WJ, Ghatan S. Congenital pterional dermal sinus in an 18-month-old child: Case report. *Neurosurgery* 2007;61:E661.
  13. Meyer DR, Lessner AM, Yeatts RP, Linberg JV. Primary temporal fossa dermoid cysts. Characterization and surgical management. *Ophthalmology* 1999;106:342-9.
  14. Neblett CR, Caram PC, Morris R. Lateral congenital dermal sinus tract associated with an intradiploic dermoid tumor. Case report. *J Neurosurg* 1970;33:103-5.
  15. Niederhagen B, Reich RH, Zentner J. Temporal dermoid with intracranial extension: Report of a case. *J Oral Maxillofac Surg* 1998;56:1352-4.
  16. Noguchi A, Shiokawa Y, Delahaw JB Jr. Surgical anatomy for orbitozygomatic approach. *No Shinkei Geka* 2010;38:703-13.
  17. Parag P, Prakash PJ, Zachariah N. Temporal dermoid-an unusual presentation. *Pediatr Surg Int* 2001;17:77-9.
  18. Prior A, Anania P, Pacetti M, Secci F, Ravegnani M, Pavanello M, *et al.* Dermoid and epidermoid cysts of scalp: Case series of 234 consecutive patients. *World Neurosurg* 2018;120:119-24.
  19. Scolozzi P, Lombardi T, Jaques B. Congenital intracranial frontotemporal dermoid cyst presenting as a cutaneous fistula. *Head Neck* 2005;27:429-32.
  20. Shehadi JA, Alorainy IA, Johnston KM. Temporal dermoid cyst with a partial dermal sinus tract. *Can J Neurol Sci* 1999;26:321-4.
  21. Vega RA, Hidayat DT, Tye GW, Fuller CE, Rhodes JL. Intradiploic dermoid cyst of the lateral frontotemporal skull: Case report and review of the literature. *Pediatr Neurosurg* 2013;49:232-5.
  22. Wells TS, Harris GJ. Orbital dermoid cyst and sinus tract presenting with acute infection. *Ophthalmic Plast Reconstr Surg* 2004;20:465-7.

**How to cite this article:** Hong S, Maruyama K, Hatanaka R, Noguchi A, Shimoyamada H, Nagane M, *et al.* Frontotemporal dermoid cyst with incomplete dermal sinus tract in an adult: A case report. *Surg Neurol Int* 2020;11:429.