Case Report

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

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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 during fetal development. It accounts for 0.04–0.7% of all intracranial tumors. Intracranial dermoid cyst tends to occur in the midline whereas scalp (extracranial) dermoid cyst may occur off-midline. 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. 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.
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 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. 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 supercili. 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, 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 (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.\(^7\)

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 et al., 1970\(^{[14]}\) | 1/M | Local swelling | R | N | A | No recurrence at 3 year |
| Green, 1973\(^{[8]}\) | 6/M | Local swelling | R | N | A | No recurrence at 9 months |
| Cullen, 1974\(^{[6]}\) | 2/F | Discharging sinuses | R | N | A | |
| Hong, 1998\(^{[1]}\) | 2/F | Discharging sinus | R | N | A | |
| Niederhagen et al., 1998\(^{[15]}\) | 1/M | Draining sinus | R | N | A | |
| Meyer et al., 1999\(^{[2]}\) | 2/M | Discharging sinus, orbital cellulitis | R | N | A | No recurrence at 8 years |
| Akita et al., 2000\(^{[2]}\) | 3/F | Discharging sinus, local infection | R | Y | A | No recurrence at 6 months |
| Parag et al., 2001\(^{[12]}\) | 1/F | Discharging sinus | L | N | A | |
| Lacey et al., 2003\(^{[11]}\) | 2/F | Discharging sinus, orbital cellulitis | L | Y | A | |
| | 5/F | Discharging sinus | L | N | A | |
| | 5/F | Discharging sinus, local swelling | L | N | A | |
| Wells and Harris, 2004\(^{[22]}\) | 0/M | Discharging sinus, orbital cellulitis | ND | N | A | No recurrence at 7 months |
| Scolozzi et al., 2005\(^{[9]}\) | 1/F | Dermal pits | R | N | A | No recurrence at 8 months |
| Mack and Ghatan, 2007\(^{[12]}\) | 1/F | Discharging sinus | L | N | A | No recurrence at 1 year |
| Barnard et al., 2012\(^{[6]}\) | 3/F | Local infection, vomiting | R | Y | A | |
| Bliss et al., 2016\(^{[4]}\) | 3/F | Discharging sinus, orbital cellulitis | R | N | A | Transient paresis of FBFN |
| Barkley et al., 2019\(^{[2]}\) | 1/F | Discharging sinus, orbital cellulitis | R | N | A | No recurrence at 1.5 months |
| Shehadi et al., 1999\(^{[20]}\) | 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 |

R: Right, L: Left, N: No, Y: Yes, ND: Not described in the abstract, FBFN: Frontal branch of the facial nerve
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. 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. MR imaging is useful if the cyst has associated sinus tract extending intracranially, 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.

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Conflicts of interest

There are no conflicts of interest.

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