

Reconstructive

CASE REPORT

A Case of Dissecting Cellulitis which Was Initially Suspected to Be a Trichilemmal Cyst

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Summary: We treat infected cysts on a daily basis, but it is difficult to diagnose similar lesions produced by inflammatory conditions that are not primarily caused by bacteria. Dissecting cellulitis of the scalp (DCS) is a chronic inflammatory disease that results in disfiguring, painful, and purulent lesions. It often takes a long time to diagnose. The pathophysiology of DCS remains unclear. Various treatments for DCS have been proposed, depending on the severity of the disease. However, none of these treatments are clearly superior to the others. If DCS spreads to the entire occipital region, aggressive surgical treatment may be beneficial in terms of the patient's quality of life. However, surgical interventions, such as drainage, are not effective at preventing the progression of the disease. Herein, we report the case of a young female patient who developed a cyst in the occipital region. We initially suspected that the lesion was a normal infected trichilemmal cyst. However, DCS was subsequently suspected because the lesion exhibited an unusual course after drainage and debridement. We consider that we made a diagnosis relatively early, but if we had sufficient knowledge about DCS we could have made a diagnosis even earlier by performing debridement sooner. Minocycline was administered for 5 months, which caused the lesion to disappear. After 2 years, no recurrence had been observed. (Plast Reconstr Surg Glob Open 2021;9:e3661; doi: 10.1097/GOX.000000000003661; Published online 6 July 2021.)

e treat infected cysts on a daily basis, but it is difficult to diagnose similar lesions produced by inflammatory conditions that are not primarily caused by bacteria. Dissecting cellulitis of the scalp (DCS) is a type of chronic inflammatory dermatosis which results in disfiguring painful and purulent lesions. It is very difficult to treat because it is resistant to standard treatments, including intralesional or topical steroids and antibiotics.^{1–3} Moreover, it often takes a long time to diagnose.¹

Herein, we report the case of a patient who developed a cyst in the occipital region. An infected cyst was initially suspected, and drainage was performed, but the lesion did not heal. Moreover, the lesion's postdebridement course was unusual, which led us to suspect DCS.

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PATIENT

In January 2018, a 25-year-old woman visited our department complaining of a painful subcutaneous swelling in the occipital region, which had first arisen 1 month earlier. She had no history of acne conglobata or hidradenitis suppurativa. We suspected that the subcutaneous swelling was a standard infected trichilemmal cyst and performed drainage on the same day.

At that time, the central sebum reservoir could not be located; therefore, an incision was made in the lower portion of the cyst so that effective drainage could be performed. About 50 cm^3 of turbid cystic fluid containing keratin was observed. The inside of the cyst exhibited poor granulation tissue formation. A small amount of *Staphylococcus capitis* was detected in a culture. The cyst gradually contracted, but 2 months later, some fistulas caused by the incision remained, and no further improvement was observed. Next, we suspected that the lesion might be related to problems with the periosteal sinus, but no such problems were found on computed tomography.

In July 2018 (6 months after the first treatment), a small fistula, in which keratin and sebum were found at the center of the lesion; several fistulas caused by the previous incision, which exhibited poor granulation; and a

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Fig. 1. Findings observed after the debridement surgery. A, An area of induration (size: 6×4 cm) remained. The cyst only shrank slightly, and several fistulas remained. B, A small fistula (which excreted keratin and sebum) was seen at the center of the cyst. C, Several fistulas caused by the previous incision, which exhibited poor granulation, and a thick scar around the lesion were removed under general anesthesia.

thick scar around the lesion were removed under general anesthesia (Fig. 1). However, 2 months later, the fistulas still remained.

Based on the assumption that the lesion had been caused by dissecting cellulitis, 50 mg/day minocycline was orally administered. No cystic fluid was observed after 1 month, and the fistula closed 3 months later. Minocycline was discontinued after 5 months, and no relapse of symptoms had been observed after 2 years (Fig. 2).

DISCUSSION

In the early literature, DCS was reported to be primarily associated with young Africans, but a recent series of cases suggests that it also occurs in Asians and Whites. The incidence rate of the condition is unclear. It exhibits a marked male predominance (80%–95%). The mean age at diagnosis is 24.9 years, and the median time between disease onset and diagnosis is 12 months (1 day–10 years).^{1–3} Our patient belonged to the predominant age group, but was a woman, which is rare.

DCS involves the development of single or multiple pustules and nodules, which fluctuate and interconnect

to form draining sinuses and tracts. The hairy scalp regions are mainly affected. The majority of patients will eventually develop scarring alopecia. The most commonly affected region is the vertex region, followed by the occipital region.^{1–3}

Immediately after a DCS lesion develops, it can become aseptic.⁴ In the current case, it may have been possible to suspect DCS earlier because we detected the lowpathogenic bacterium *Staphylococcus capitis*. However, we only suspected DCS after performing thorough surgery, as inflammation can be prolonged by the presence of a residual cyst wall; therefore, earlier surgery may have been diagnostically useful. In this case, we were able to observe the early stages of DCS, and performing drainage immediately after cyst formation did not cause alopecia.

The pathophysiology of DCS remains unclear. Along with pilonidal cysts, hidradenitis suppurativa, and acne conglobata, DCS is part of the "follicular occlusion tetrad," suggesting a common pathogenesis involving deep follicular occlusion and infection.¹⁻³ In these conditions, the loss of immune tolerance to alloantigens in the hair follicle may lead to an inflammatory reaction.⁵ The



Fig. 2. Findings obtained before and after minocycline treatment. A, Even 2 months after the debridement, fistulas remained, although the cyst had shrunk further. B, An oral dose of 50 mg/day minocycline was administered. No drainage fluid was observed after 1 month, and the fistula had closed 3 months later.

amelioration of the lesion seen after antibiotic treatment favors this hypothesis. An initial trauma could also cause a break in the skin barrier. In the current case, the lesion formed in the occipital region, which is the weight-bearing region during sleep, and hence, may be susceptible to pressure.¹

DCS is difficult to treat. Various treatments have been proposed, depending on the severity of the disease.² However, none of the treatment options are clearly superior to the others.³

It has been reported that to alleviate pain, surgical interventions, such as drainage, should be performed, but this approach does not prevent progression.¹ If DCS has spread to the entire occipital region, aggressive surgical treatment may be helpful in terms of patients' quality of life.⁶

As for nonsurgical treatments, isotretinoin, corticosteroids, doxycycline, minocycline, and adalimumab have all been employed.³ However, all of these drugs are considered unsuitable for young women, as they can be pregnant. Our patient's symptoms were considered to be mild, and the use of minocycline and doxycycline in such cases has been advocated.^{2,7} Minocycline is widely used to treat acne (at a dose of 50–100 mg/day). Although its side effects include interstitial pneumonia and liver dysfunction, they are considered to be relatively rare; therefore, we chose to use it.

Minocycline has antiinflammatory effects; for example, it can inhibit lipase activity, leukocyte migration, and active oxygen activity, in addition to antibacterial effects.^{8,9} Inhibiting lipase activity suppresses the production of free fatty acids from sebum by bacteria. Although there have been some reported cases of DCS in which minocycline was not effective,^{8,9} it was effective in the present case, and no relapse subsequently occurred.

CONCLUSIONS

We initially suspected that an occipital cyst that arose in a young female patient was a standard infected

trichilemmal cyst. DCS was subsequently suspected because the lesion exhibited an unusual course after drainage and debridement. We consider that we made a diagnosis relatively early, but if we had sufficient knowledge about DCS, we could have made a diagnosis earlier by performing debridement sooner. After minocycline was administered for 5 months, the lesion disappeared. After 2 years, no recurrence had been observed.

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