Case Report

Iraqi National Journal of Medicine. July 2025, Volume 7, Issue 2

Central cervical cleft: A very rare case of cervical congenital malformation

Wael J. Alshihaby

Basra Teaching Hospital, Basra Cochlear Implant Center, Basra, Iraq

ABSTRACT

Central cervical cleft is an rare congenital midline anomaly of the neck, typically presenting as a vertically oriented skin defect accompanied by a subcutaneous fibrous band and, in many cases, an associated skin tag. This case report presents a case involving a five-year-old female who exhibited a midline cervical lesion with intermittent clear discharge but no associated pain or functional impairment. Clinical examination identified a characteristic cutaneous defect with a prominent skin tag in the midline of the anterior neck. Radiological evaluation, including ultrasound and CT scan, confirmed the localized nature of the anomaly and excluded deeper structural involvement. After appropriate counseling and reassurance of the benign nature of the lesion, surgical management was undertaken under general anesthesia. The excision was followed by multilayered wound closure incorporating multiple Z-plasty incisions to optimize healing and minimize scar formation. The postoperative course was uneventful, and the cosmetic outcome was further enhanced through regular follow-up and scar management using silicone gel. This case highlights the importance of early recognition and surgical correction of central cervical cleft to prevent potential functional limitations and achieve superior aesthetic results. Z-plasty remains a valuable reconstructive technique in such congenital neck anomalies, particularly in pediatric patients.

Keywords: central cervical cleft, congenital neck anomaly, Z-plasty, pediatric surgery, cosmetic reconstruction

Corresponding author: Wael J. Alshihaby. E-mail: alshihaby@iamrs.edu.iq
Disclaimer: The author has no conflict of interest.

DOI: https://doi.org/10.37319/iqnjm.7.2.18

Received: 18 APR 2025 Accepted: 25 MAY 2025 Published online: 15 JUL 2025

INTRODUCTION

The first description of congenital midline cervical cleft (CMCC) was by the German anatomist Luschka in 1848[1], with the first detailed report published by Bailey in 1924. However, the anomaly was not fully described until Ombredanne's work in 1946. [2-4] It arises from defective midline fusion of the first and second branchial arches.[5] Clinically, it presents as a midline cervical skin defect with an underlying fibrous band and a skin tag. Unlike thyroglossal duct cysts or dermoid cysts, it lacks an epithelial-lined tract [6] If left untreated, the lesion may lead to progressive contracture and limitation of neck extension.[7]

Surgical excision with reconstruction remains the gold standard treatment to prevent long-term functional and cosmetic concerns.[6] Thus, in cases of central cervical cleft, early surgical intervention is advised to prevent significant scarring and functional impairment.

CASE PRESENTATION

A five-year-old female presented to my private Ear, Nose & Throat ENT clinic with a congenital midline cervical anomaly. The lesion had been present since birth. Although the child experienced no pain or respiratory distress, intermittent clear discharge from the lower part of the cleft was reported. The parents sought surgical intervention primarily for cosmetic reasons before school enrollment.

On clinical examination, a vertically oriented midline defect was observed, extending from the suprasternal notch to the mid-neck. A firm, hairy, skin-colored tag was present at the superior end of the cleft. The surrounding skin appeared normal, with no signs of infection or tenderness. No palpable masses or underlying sinus tracts were detected. (Fig. 1)

Based on the characteristic presentation, a clinical diagnosis of central cervical cleft was made, and surgical excision with Z-plasty reconstruction was planned.

Ultrasonographic examination of the neck was conducted to evaluate any deeper extensions of the cleft and its relationship to vital neck structures. The scan revealed no extension into deeper tissues and no involvement with the carotid vessels, jugular veins, or any neural structures. The cleft was confined to the superficial layers of the neck. A CT scan further confirmed these findings. Although a small artery was noted at the lower end of the cleft, it was not considered surgically significant

Histopathological analysis of the excised cleft and the associated skin tag revealed a fibrous tissue-lined cleft with an overlying hairy skin tag, consistent with the diagnosis of a central cervical cleft (Fig. 2).

For surgical management, the patient was placed in a supine position on the operating table, with a sandwich pillow placed under the shoulders to allow for a fully extended neck. This positioning facilitated optimal exposure of the surgical site and ensured precise incision placement (Fig. 3).

Under general anesthesia and cuffed endotracheal intubation, the surgical procedure was carried out.

The cleft was marked along its vertical midline, with careful delineation of the skin tag at its inferior aspect (Fig. 4). The cleft and the associated skin tag were excised using precise dissection techniques. Z-Plasty Reconstruction: Multiple Z-plasty incisions were designed and executed to reduce wound tension and improve the cosmetic outcome (Fig. 5). Layered Closure: Deep layers: 4-0 Polydioxanone PDS sutures were used to approximate subcutaneous tissues. Skin closure: Interrupted 5-0 nylon sutures were used for primary skin closure. The wound was closed carefully, and tension-reducing techniques were employed to minimize scarring. The patient tolerated the procedure well with no intraoperative complications.

The patient was discharged on postoperative day 2 after confirming the absence of hematoma or respiratory distress. No dressing was applied, and fusidic acid ointment was used daily. Nylon sutures were removed on postoperative day 7 after confirming adequate wound healing. The wound was cleaned with 1% Betadine solution and fusidic acid ointment. A silicone-based gel was applied for two months after wound healing complete (3weeks after surgery) to enhance the final cosmetic result. Six months later, the wound had healed well, with minimal scarring and no functional impairment (Fig. 6).

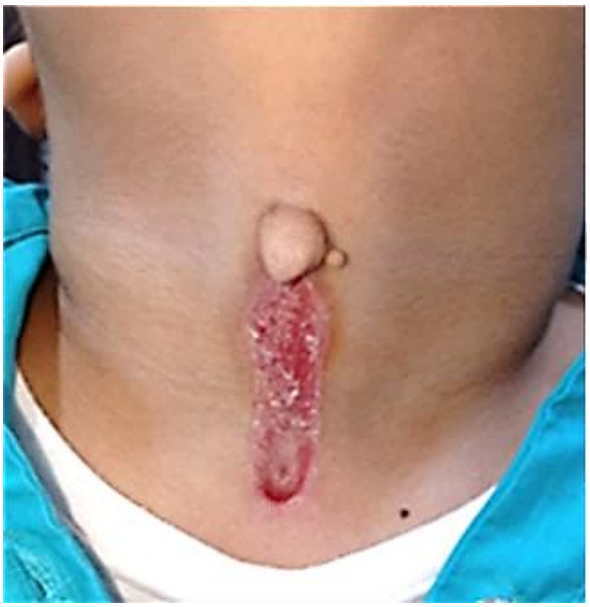

Figure 1: The congenial central cervical cleft appears as a skin defect lined with fibrous crustating epithelium with skin tag.

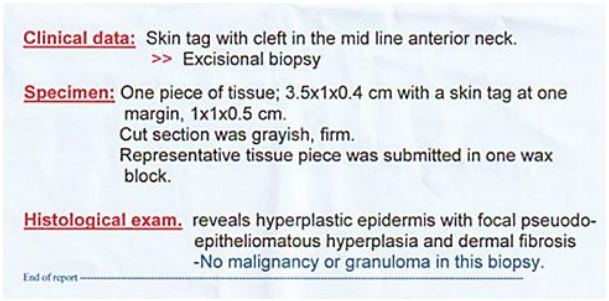

Figure 2: Histopathological report showing hyperplastic epidermis with focal pseudoepithliomatous hyperplasia and dermal fibrosis.

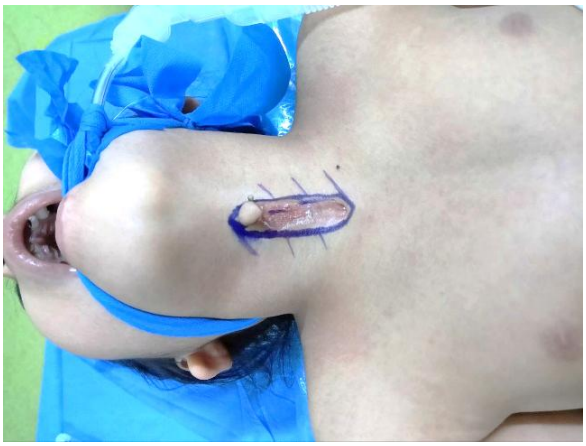

Figure 3: The patient was placed in a supine position with the head extended fully using a pillow under the shoulders to completely expose the surgical site.

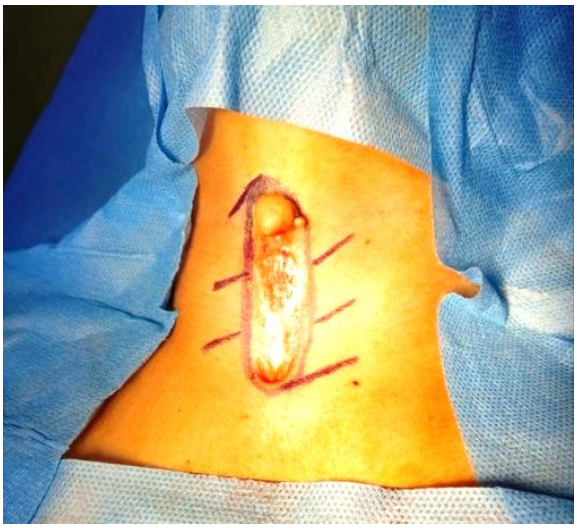

Figure 4: Skin incisions marked with a pen, followed by outlining of multiple Z-plasty flaps.

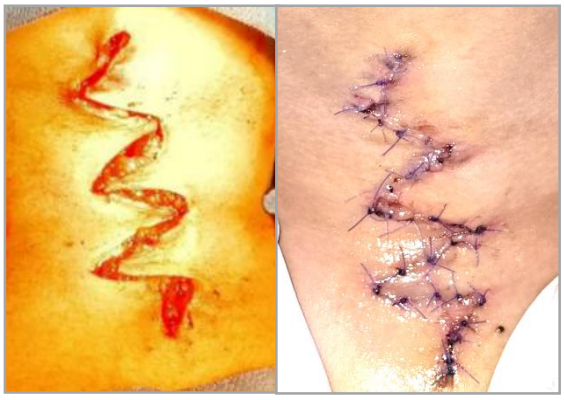

Figure 5: Multiple Z-plasty flaps approximated with no tension.

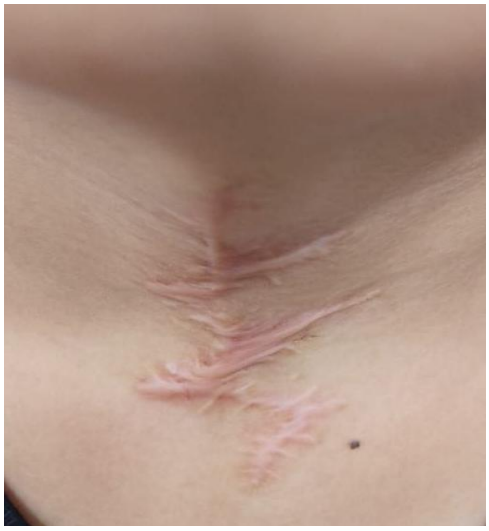

Figure 6: 6 months after suture removal, the wound shows good healing and a cosmetically acceptable primary scar.

DISCUSSION

The central cervical cleft is believed to arise from incomplete fusion of the first and second branchial arches during embryogenesis.[8] This results in defective midline skin formation and an associated subcutaneous fibrous band (Fig. 7).

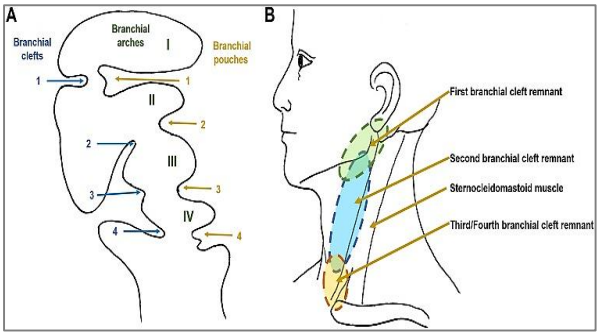

Figure 7: Failed fusion of the first and second branchial arches during embryogenic development is thought to cause congenital cervical cleft.[1]

Several case reports have highlighted successful management strategies for this rare congenital midline neck anomaly. Maschka et al. reported a similar case in an 18-month-old girl, where Z-plasty was used successfully to reconstruct the defect.[9] Similarly, Mlynarek et al. presented an extensive review and clinical report of a patient with central cervical cleft, emphasizing the importance of complete excision and tailored surgical planning.[10] D'Souza et al. described a technique involving complete excision of the fibrous cord and use of double Z-plasty flap to create a

tension-free closure and restore contour to the anterior neck.[11]

Surgical techniques for this condition vary based on the size and severity of the defect. Smaller clefts may be amenable to primary linear closure; however, this approach carries a risk of postoperative contracture and limited neck extension.[6,7] Z-plasty is widely favored for larger defects due to its ability to realign the scar and reduce the likelihood of contracture.[9-11] In more extensive cases, local flap reconstruction may be necessary to provide adequate tissue coverage and achieve optimal aesthetic and functional results.[6,7]

CONCLUSIONS

Awareness of central cervical cleft—a rare congenital anomaly—is crucial for early recognition and surgical management, which are essential to prevent long-term functional impairment and cosmetic deformity. Z-plasty remains the preferred technique, as it minimizes scar contracture and improves aesthetic outcomes.

REFERENCES

1. Von Luschka H. Ueber fistula colli congenita. Archiv für Physiologische Heilkunde. 1848;7:24–27
2. Dermawan JK, Chute DJ. Educational Case: Developmental Neck Masses and Other Neck Tumors. Academic Pathology. 2019;6. doi:10.1177/2374289519888735
3. Bailey H. Thyroglossal cysts and fistulæ. British Journal of Surgery. 1925;12(47):579–589. doi: 10.1002/bjs.1800124716.
4. Ombredanne L., Precis Clinique et Operatoire de Chirurgie Infantile, 1949, 5th edition, Masson, Paris, France.
5. Foley DS, Fallat ME. Thyroglossal duct and other congenital midline cervical anomalies. Semin Pediatr Surg. 2006;15(2):70–5.
6. McInnes CW, Benson AD, Verchere CG, Ludemann JP, Arneja JS. Management of congenital midline cervical cleft. J Craniofac Surg. 2012;23(1):e36–8.
7. Magalhães R, Louro M, Forny D, Sá Á, Franco D. Congenital midline cervical cleft: management of a case series and literature review. J Plast Reconstr Aesthet Surg. 2024;93:117–26.
8. Puscas L. Midline cervical cleft: review of an uncommon entity. Int J Pediatr. 2015;2015(1):209418.
9. Maschka DA, Clemons JE, Janis JF. Congenital midline cervical cleft case report and review. Ann Otol Rhinol Laryngol. 1995;104(10):808–11.
10. Mlynarek A, Hagr A, Tewfik TL, Nguyen VH. Congenital mid-line cervical cleft: case report and review of literature. Int J Pediatr Otorhinolaryngol. 2003;67(11):1243–9.
11. D'Souza JN, Valika T, Maddalozzo J. Surgical management of midline cervical cleft. Int J Pediatr Otorhinolaryngol. 2019;127:109657.