P4-7

Intraosseous Venous Malformation of the Zygoma: Cases Report and Literature Review

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Objective

- To summarize the clinical and radiological features and review the treatment modalities of zygomatic intraosseous venous malformations (IVMs) by reporting 4 cases and analyze the features of them together with other previous case reports.
- To help clinicians and radiologists to get a correct preoperative diagnosis and proceed with the correct treatment of zygomatic IVMs.

Cases Report

- We report 4 cases with pathologically diagnosed zygomatic IVMs, their information is shown in **table 1**. The commonalities of them were:
  - **Similar chief complaint**: a solid mass fixed to the midface which made cosmetic deformity, without any symptom, or just with slight pain.
  - **Physical and auxiliary examination:** asymmetric; a firm and solid mass protruded from the zygomatic bone. The typical CT imaging is shown in **figure 1.**

No.	Age	Sex	Pain	symptoms/ appearance	main imaging	round-shaped	internal pattern	well-defined	intact cortex	Manage-ment	Bleeding	Follow-up dysfunction or recurrence
1	35	F	Y	firm, fixed; enlarged after 2 years with pain	CBCT	Y	Dendritic	Y	N	En-bloc excision	N	2 yr, N
2	57	M	N	firm, fixed	CT	Y	Spongious	Y	N	En-bloc excision	N	1 yr, N
3	33	F	Y	firm, fixed; pain during menstruation but painless of palpation.	CT	Y	Radiated	Y	N	En-bloc excision	N	1 yr, N
4	54	M	N	firm, fixed	CT	Y	Honeycomb	Y	N	En-bloc excision	N	N

Table 1. Summary of demographic, clinical, radiographic, and histopathologic findings of present 4 cases. F = female; M = male; CBCT = cone-beam computed tomography; CT = computed tomography; N = Np; yr = year(s).

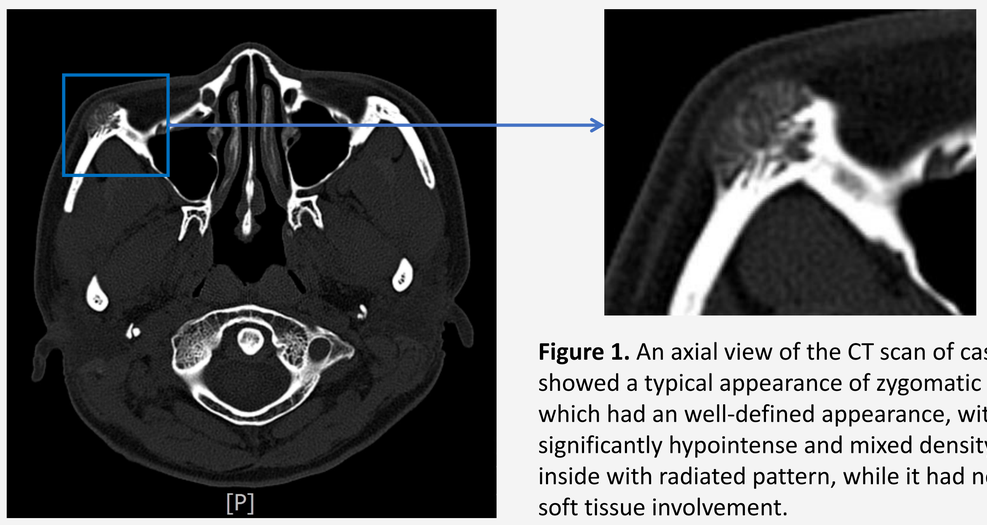

Figure 1. An axial view of the CT scan of case 3 showed a typical appearance of zygomatic IVM, which had an well-defined appearance, with significantly hypointense and mixed density inside with radiated pattern, while it had no soft tissue involvement.

Literature review

- Other 58 cases of zygomatic IVMs were reviewed and identified, according to the latest ISSVA classification of vascular anomalies.[1] (ISSVA = the International Society for the Study of Vascular Anomalies)
- **Demographic (figure 2): The ratio of female and male is 2.1 : 1 (42 : 20),** females or people in their fourth to sixth decades have a higher proportion.
- **Etiology**: unknown. Local stimulation like injury, infection, hormonal fluctuations seems to be the main reason.[2]
- **Main clinical symptoms (Figure 3):** cosmetic deformity (60/62), pain (20/62) and ocular Involvement (9/62).

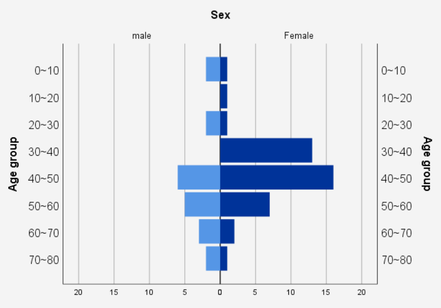

Figure 2 The population pyramid count age group by sex of zygomatic IVMs.

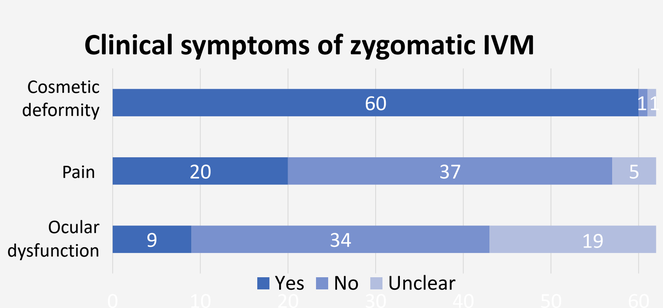

Figure 3 The first three clinical symptoms of zygomatic IVMs.

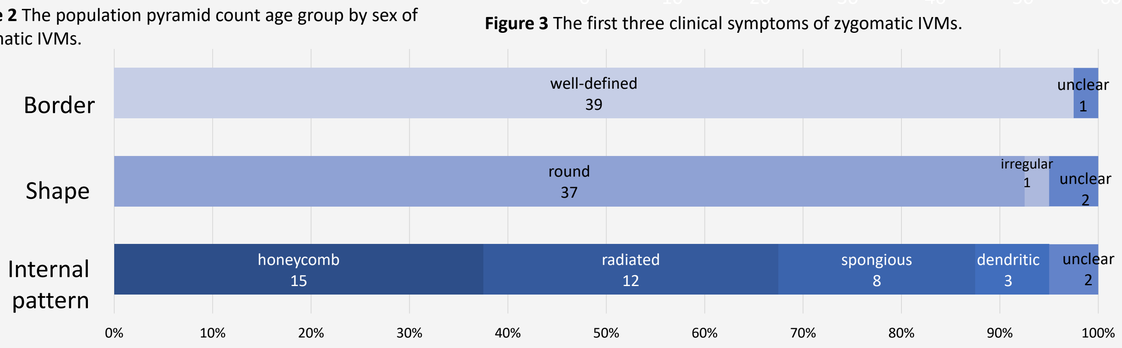

Figure 4. CT features of zygomatic IVMs and the proportion of each type.

Radiological Analysis

- Computed tomography (CT) was the most common imaging examination.
- 40 cases (included our 4 cases) which provided CT images were analyzed.
- Zygomatic IVMs usually have a well-defined border and a round shape appearance, protruding to the contour of zygoma.
- Different internal patterns were identified and categorized into four types: honeycomb, radiated, spongious and dendritic. **(Figure 4-5)**
  - Honeycomb: multiple hyperintense foci (dots) inside.
  - Radiated: trabecula radiating from a center like sunburst or spoke wheel.
  - Dendritic: trabecula distributed without a center.
  - Spongious: A grey background filled with small cavities like sponge.

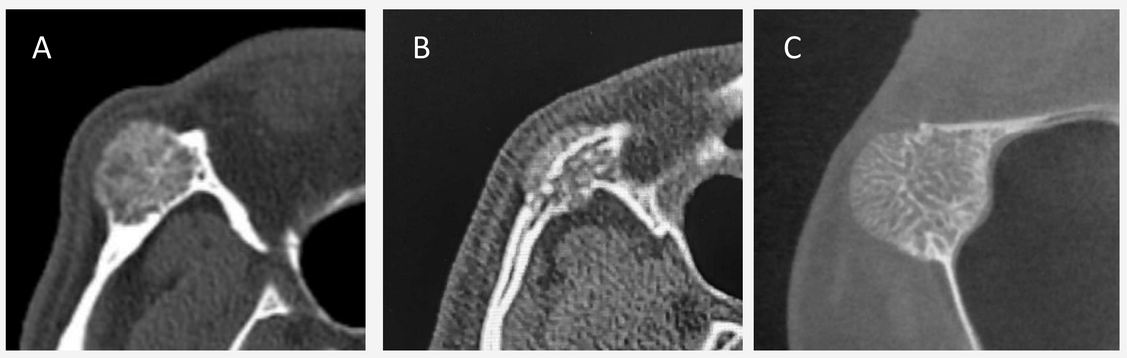

Figure 5. Different internal patterns of zygomatic IVM in CT or CBCT scan:
(A) spongious pattern (case 2); (B) honeycomb pattern (case 4). (C) dendritic pattern (case 1)

Treatment and Histopathology

- Different methods of treatment was reported with different events and prognosis. **(Figure 6) En bloc Excision** is likely to be the best option, with little risk of hemorrhage and recurrence.
- **Incisional biopsy: Cautious!** High risk of bleeding.
- **Reconstruction:** Different materials were reported and applicable. (Figure 7) CAD/CAM was used in some cases.
- **Histopathology: (figure 8)**
  - proliferative, dilated and vascular lined by flattened endothelial cells;
  - trabeculae absorption.

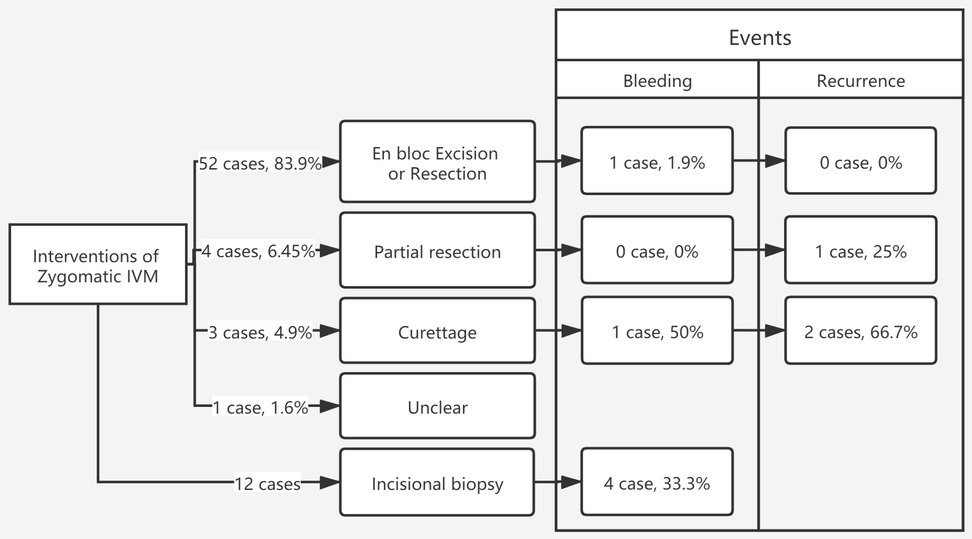

Figure 6. Different interventions, bleeding incidence, and prognosis of zygomatic IVM.

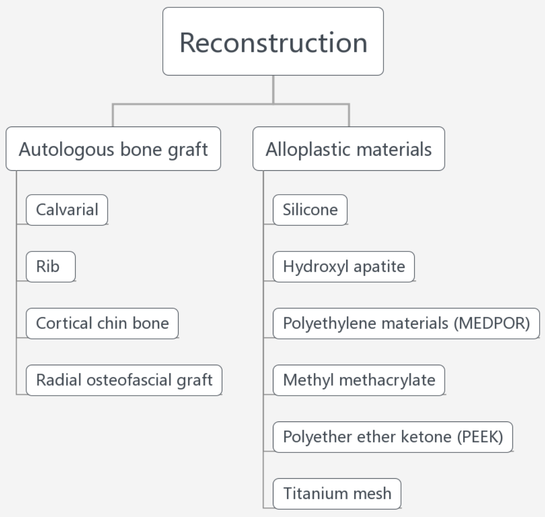

Figure 7. The options of reconstruction reported in previous cases.

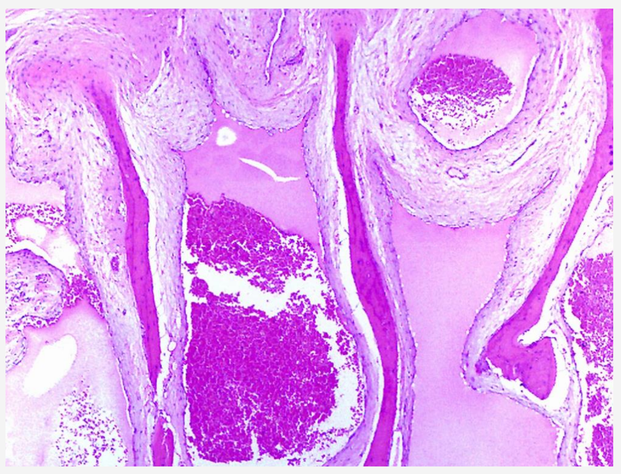

Figure 8. The histological appearance of case 3 (under 100x magnification).

Conclusions

- Zygomatic IVMs were rare but was the most common vascular malformation of zygoma.
- There clinical and radiological features was established in this study to help to obtain a correct diagnosis.

References

[1]Monroe EJ. Brief Description of ISSVA Classification for Radiologists. *Tech Vasc Interv Radiol*. 2019;22(4):100628. doi:10.1016/j.tvir.2019.100628

[2]Fábián Z, Szabó G, Petrovan C, et al. Intraosseous venous malformation of the zygomatico-orbital complex. Case report and literature review with focus on confusions in vascular lesion terms. Oral Maxillofac Surg. 2018;22(2):241-247. doi:10.1007/s10006-018-0691-0